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OBSERVATIONS ON VISCERAL PAIN*

(1) Visceral Tenderness

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The many and diverse views that have been held on the subject of visceral sensation are due chiefly to differences of observation, which lead naturally to differences in interpretation. So long as the basic clinical and experimental findings remain in dispute there can be no agreement about the sensory mechanisms involved in visceral pain. The actual situation of the deep tenderness that occurs in abdominal disease; the presence and distribution of skin hyperesthesia in visceral disorders; the effect of procaine when injected into the area of pain-reference: these are all examples of conditions in which directly opposing observations have been made.

It is necessary at the present time to re-examine some of the facts of visceral sensation and to report simply the findings. In this paper are described experiments on the location of deep tenderness found in patients suffering from chronic peptic ulcer. By tenderness is meant that a sensation of pain is evoked when the area is pressed upon. The questions that the experiments were designed to answer are:

In patients suffering from chronic peptic ulcer,

(I) Is the ulcerated wall of the stomach or duodenum itself tender (as claimed by Kinsella and Palmer)?

or

(II) is the pain that is produced by pressure in the epigastrium invariably due to irritation of the sensitive anterior parietal peritoneum by the ulcerated viscus (as suggested by Morley and Twining)?

or

(III) does the pain arise from pressure on hyperesthetic areas in the anterior abdominal wall caused by a viscero-sensory radiation from the ulcerated area of the stomach or duodenum (the classical Mackenzie view)?

(I) EPIGASTRIC TENDERNESS IN PATIENTS WITH CHRONIC PEPTIC ULCER WAS ABOLISHED WHEN THE SPLANCHNIC NERVES WERE BLOCKED WITH PROCAINE

Technic of Splanchnic Block

No sedative was given. The abdomen was examined and then, with the patient in the sitting position, 25 ml. of 1 per cent procaine were injected about

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the anterolateral aspect of the body of the first lumbar vertebra on each side. The injection was accepted as satisfactory only when there was faintness and a fall of blood pressure. The patient was laid down again, and the abdomen re-examined. Evidence of skin anesthesia was looked for, lest the 12th intercostal or the 1st lumbar nerves should have been blocked. Anesthesia was not found in any case.

In the first two patients reported below, 50 ml. of 1 per cent procaine were injected into the muscles of the lumbar region before splanchnic block was attempted. This injection resulted in no alteration in the epigastric tenderness. It was not thought necessary to carry out further controls on the effect of this amount of procaine on pain sensitivity.

CASE REPORTS

Case 1.—C. P. Male. Age 54. History of 17 years ulcer pain. A large pyloric ulcer was demonstrated radiologically. Examined during an acute exacerbation. Patient ill and frail.

Examination: (a) No hyperesthesia of the skin of the epigastrium; (b) marked tenderness on moderate pressure in the epigastrium. Tenderness diffuse, but maximal at one place (x, Fig. 1).

SPLANCHNIC BLOCK

Re-Examination: All tenderness gone. Now possible to palpate deeply in the upper abdomen with no trace of discomfort.

At operation one week later, a large ulcer of the pylorus and first part of the duodenum was found.

Case 2.—G. C. Male. Age 45. History of 5 years ulcer pain. Duodenal ulcer demonstrated radiologically. Examined during an acute exacerbation.

Examination: (a) No hyperesthesia of the skin of the epigastrium; (b) Marked tenderness on moderate pressure high up in the epigastrium. Tenderness well localized to one place (x, Fig. 2).

SPLANCHNIC BLOCK

Re-Examination: All tenderness gone. Now possible to palpate deeply in the upper abdomen with no trace of discomfort.

At Operation 9 days later, a large ulcer of the anterior wall of the first part of the duodenum was found.

Case 3.—B. D. Female. Age 50. History of 10 years ulcer pain. Duodenal ulcer demonstrated radiologically.

Examination: (a) No hyperesthesia of the skin of the epigastrium; (b) Deep tenderness in the right epigastrium, well localized to one place (x, Fig. 3).

SPLANCHNIC BLOCK

Re-Examination: All tenderness gone. Now possible to palpate deeply in the upper abdomen with no trace of discomfort.

At Operation 5 days later, an ulcer involving the anterior wall of the first part of the duodenum was found.

Case 4.—R. M. Male. Age 40. History of 7 years ulcer pain. Duodenal ulcer demonstrated radiologically. Examined at the end of an acute exacerbation.

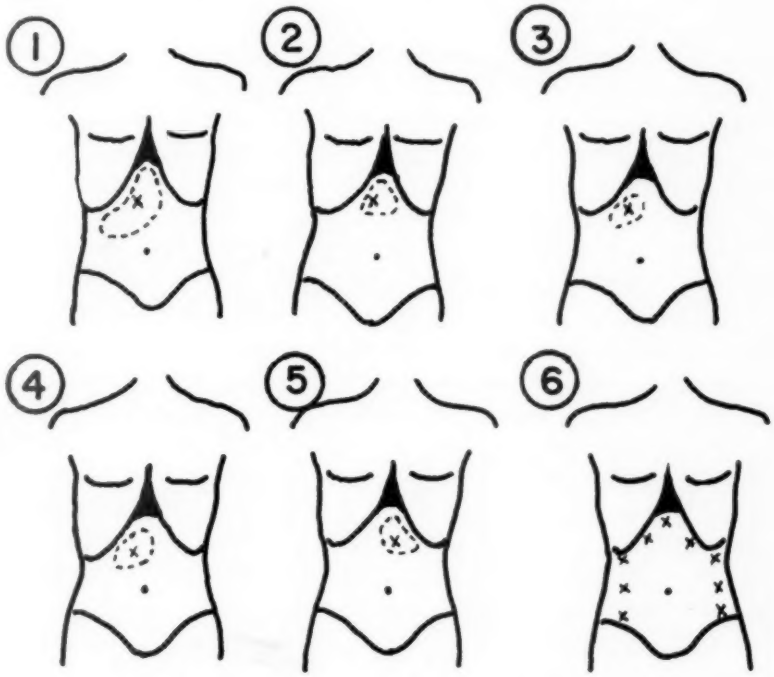
Examination: (a) No hyperesthesia of the skin of the epigastrium. (b) Deep tenderness in the right epigastrium, well localized to one place (x, Fig. 4).

SPLANCHNIC BLOCK

Re-Examination: All tenderness gone. Now possible to palpate deeply in the upper abdomen with no trace of discomfort.

Patient not brought to operation.

Case 5.—J. G. P. Male. Age 40. History of 10 years ulcer pain. Gastric ulcer demonstrated radiologically. Examined in an acute exacerbation.



FIGS. 1–5.—Site of maximal tenderness in each case. (Cases 1–5.)
FIG. 6.—Sites of injection of procaine to produce abdominal wall block.

Examination: (a) No hyperesthesia of the skin of the epigastrium; (b) Marked tenderness on moderate pressure in the left epigastrium. Maximal at one place (x, Fig. 5).

SPLANCHNIC BLOCK

Re-Examination: All tenderness gone. Now possible to palpate deeply in the upper abdomen with no trace of discomfort.

At Operation 4 days later, an ulcer of the lesser curve of the stomach was found, forming a mass about ¾" in diameter, free from adhesions in front and behind.

CONCLUSIONS

Epigastric tenderness occurring in patients suffering from chronic peptic ulcer was abolished when the splanchnic nerves were anesthetised. The peritoneal nerve supply was still intact; the tenderness could not have been due, therefore, to irritation of the anterior parietal peritoneum by the ulcerated

viscus. The results indicated either that the ulcerated area was itself the site of the deep tenderness; or that the tenderness was due to pressure on hypersensitive areas in the abdominal wall, for the afferent limb of any viscerosensory radiation would have been abolished by the splanchnic block.

In order to investigate the pain sensitivity of the ulcerated viscus it was necessary to stimulate the ulcer area directly, and not through the sensitive anterior body wall.

(II) THE WALL OF THE STOMACH OR DUODENUM INVOLVED IN AN ULCER WAS SENSITIVE TO DIRECT MECHANICAL STIMULATION

Operation was performed under abdominal wall block on patients suffering from chronic peptic ulcer and in whom epigastric tenderness had been an obvious clinical feature. The anesthetic was introduced by subcostal injection and into the rectus sheath (Fig. 6). A total of 150 ml. of 1 per cent procaine, containing a concentration of 1 in 100,000 adrenaline hydrochloride, was employed. As the observations were on the persistence and not on the abolition of pain, controls were not needed.

The patients were given ⅙ gr. morphia or of "Omnopon" as a sedative. This did not affect the precision of the pain responses, although they became unreliable if heavier sedation was employed. The abdomen was opened through a right upper paramedian incision.

There are many difficulties in making direct observations on visceral sensation in the conscious individual. The patient needs to be fully responsive to unpleasant stimuli, and yet should lie tranquil and calm in order that the response to any stimulation of the viscus may be precise and significant. If the patient is in discomfort it is impossible to rely on his appreciation of a new pain. In addition, observations have to be carried out with much gentleness, and the stimulus applied to the desired area without causing any interference with other tissues, as, for example, by deforming or pulling on the lesser omentum.

These conditions are not easy to fulfill, and only when they were met was the experiment accepted. In some patients it was possible to make simple observations. In others, of more phlegmatic mould, the observations could be extended.

Case 6.—J. D. Male. Age 40. Gastric ulcer. Preoperative tenderness in left epigastrium.

Operation: Gentle retraction of the left half of the wound revealed an ulcer at the centre of the lesser curve, forming a mass in the stomach wall about ¾ inch in diameter. The ulcer mass extended slightly into the lesser omentum. It was free from adhesions in front and behind.

Tests. (a) Ulcer mass gently pressed with finger—***continuous pain while pressure maintained;*** (b) Insertion of a fine hypodermic needle into the mass—***immediate pain as needle introduced;*** (c) Injection of 0.5 ml. 6 per cent NaCl into the mass—***pain, lasting several minutes.***

All these tests were performed away from the lesser omentum in that part of the mass in the anterior stomach wall.

(d) Similar tests to (a) (b) and (c) were carried out at a normal part of the lesser curve—no sensation.

General anesthetic given. Partial gastrectomy performed.

Case 7.—B. D. Female. Age 50. Duodenal ulcer. Preoperative tenderness in the right epigastrium.

Operation: Scarred first part of duodenum with ulcer mass ¾ inch in diameter adherent to the pancreas behind, but free from adhesions in front.

Tests: (a) Anterior surface of the mass gently rubbed with finger—***continuous pain;*** (b) Anterior surface of the mass gently pressed with the finger—***continuous pain.***

General anesthetic given. Partial gastrectomy performed.

Case 8.—E. K. R. Female. Age 59. Gastric ulcer. Preoperative tenderness in the left epigastrium.

Operation: Gentle retraction of the left half of the wound revealed an ulcer at the lower part of the lesser curve, forming a mass in the stomach wall about 1 inch in diameter, not extending into the lesser omentum. It was free from adhesions in front and behind.

Tests: (a) Ulcer mass gently rubbed with finger—***continuous pain;*** (b) Ulcer mass gently pressed with finger—***continuous pain;*** (c) Abdominal wall depressed against ulcer mass—***pain*** (***as preoperatively***); (d) Anterior parietal peritoneum, where it was seen to impinge against the ulcer, rubbed with the finger—***no sensation*** (***confirming the abdominal wall anesthesia***); (e) Left hand introduced between the stomach and the anterior abdominal wall, and the abdominal wall pressed against the hand—***no sensation;*** (f) Hand removed and abdominal wall pressed against the ulcer mass—***pain as before;*** (g) Needle inserted into the ulcer mass—***immediate pain as needle introduced;*** (h) Injection of 0.5 ml. 6 per cent NaCl into the mass—***pain, lasting several minutes;*** (i) Pressure on the stomach, rubbing with the finger, insertion of needle and injection of 6 per cent NaCl at a normal part of the lesser curve—***no sensation.***

General anesthetic given. Partial gastrectomy performed.

Case 9.—J. P. Male. Age 37. Duodenal ulcer. Preoperative tenderness in the right epigastrium.

Operation: Ulcer mass 1½ inches in diameter occupying the first part of the duodenum and adherent to the pancreas, but free from adhesions in front.

Tests: (a) Anterior surface of the mass gently rubbed with finger—***continuous pain;*** (b) Injection of 0.5 ml. 6 per cent NaCl into the mass in the anterior wall of the duodenum—***pain, lasting several minutes;*** (c) Injection repeated at normal point on the lesser curve—***no sensation.***

General anesthetic given. Partial gastrectomy performed.

Case 10.—G. W. E. Male. Age 48. Gastric ulcer. Preoperative tenderness in the centre of the epigastrium.

Operation: Ulcer at the centre of the lesser curve, forming a mass 1 inch in diameter. The lesser omentum was drawn into the edge of the lump, but no extension of the mass into the omentum. Mass free from adhesions in front and behind.

Tests: (a) Anterior surface of the mass gently rubbed with finger—***continuous pain;*** (b) Insertion of a fine hypodermic needle into the anterior aspect of the mass—***patient instantly said "Oh! you're sticking a needle in me!" This point was ½ inch from the lesser curve border of the stomach, and the needle entered about ¼ inch in depth;*** (c) Injection of 0.5 ml. 6 per cent NaCl into the mass—***pain, lasting several minutes;*** (d) Pressure on the stomach, rubbing with the finger, insertion of a needle and injection of 6 per cent NaCl at a normal point of the lesser curve—***no sensation.***

General anesthetic given. Partial gastrectomy performed.

CONCLUSION

Direct stimulation of the exposed viscus showed that the stomach or duodenal wall involved in an ulcer was sensitive to pressure, to needle prick, and to chemical stimulation. This pain sensitivity of the viscus could not be due to pressure on hypersensitive nerves in the body wall or to irritation of the anterior parietal peritoneum. The ulcerated viscus itself was sensitive to mechanical and chemical stimulation.

DISCUSSION

There are two afferent pathways from the upper abdomen. One by the splanchnic nerves, the other by the nerves of the abdominal wall.

When the splanchnic nerves were blocked and the abdominal wall nerves were intact, the epigastric tenderness associated with a peptic ulcer was found to be abolished.

When the abdominal wall nerves were blocked, but the splanchnic nerves were intact, the exposed ulcerated area of the stomach or duodenum was found to be sensitive to pressure, to pin prick, and to chemical stimulation. The non-ulcerated part of the lesser curve of the stomach was not sensitive to these stimuli.

It is possible, therefore, to conclude that abdominal tenderness associated with a peptic ulcer is produced by pressure on the ulcerated wall of the stomach or duodenum. *That is, pain arises directly from the viscus.* The afferent pathway is by the splanchnic nerves. This observation is in agreement with the recent findings of Ray and Neill, who have confirmed by valuable studies in sympathectomised patients that pain sensitivity from the stomach is mediated wholly by afferents running with the visceral nerves.

This conclusion does not, of course, deny that if the anterior parietal peritoneum is irritated by a large or inflamed ulcer, local tenderness will result. It is rather to emphasize that tenderness can occur from stimulation of the ulcerated viscus itself.

The present experiments do not demonstrate in which part of the ulcerated stomach wall the pain sensitivity resides. They do show, however, that the pain sensitivity is within the wall of the viscus, and is not dependent on the involvement of the parietal peritoneum, the lesser omentum, or the retroperitoneal tissues.

There is no need, therefore, to postulate a viscero-sensory radiation to explain the epigastric tenderness in cases of peptic ulcer, but the presence of such a radiation is not disproved by the experiments. The viscero-sensory radiation has for long been a theoretical conception, based largely on the inability to explain visceral tenderness in any other way, for the diseased viscus was believed to be insensitive to painful stimulation. Although the present results show that the ulcerated wall of stomach or duodenum is itself tender, it could be argued that in addition to the direct visceral tenderness there was also body wall tenderness, due to a viscero-sensory radiation. The absence of hyperalgesia in the epigastrium does not support this view. Never-

theless, unless the patient is able to estimate whether the deep tenderness is diminished (or not) by abdominal wall anesthesia, there is no certain demonstration of the presence or absence of a viscero-sensory radiation. Such measurement of the degree of pain introduces difficult subjective problems, and needs to be the subject of a separate investigation.

SUMMARY

I. In patients with chronic peptic ulcer,
(i) the epigastric tenderness was abolished by splanchnic block,
(ii) the exposed ulcerated stomach or duodenum was sensitive to pressure, to pin prick, and to chemical stimulation. The non-ulcerated stomach wall was insensitive to these stimuli.

II. It is concluded that the epigastric tenderness was not due to irritation of the anterior parietal peritoneum by the ulcer, but to pressure on the viscus itself. That is, the pain arose directly within the ulcerated viscus.

III. There was no evidence of a viscero-sensory radiation, but such a radiation is not disproved by these experiments.

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HEMOPHILIA*

PROBLEM OF SURGICAL INTERVENTION FOR ACCOMPANYING DISEASES REVIEW OF THE LITERATURE AND REPORT OF A CASE

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THE NUMBER OF CASES OF HEMOPHILIA who have undergone minor surgical procedures is very great and will not be a subject of review in this report. However, Birch[1] in a monograph on hemophilia in 1937 reported the following in regard to surgery in 113 cases of hemophilia studied. Analysis of the cause of death showed that the greatest number were due to hemorrhage following surgical procedures, all but one of which were operations of a minor nature. There can be no doubt that the great majority of hemophiliacs who undergo minor surgical procedures at the present time probably are brought through safely to recovery. Nevertheless, even the simplest operation still presents a serious problem calling for intensive treatment with prophylactic transfusions or injections of fresh plasma or Fraction I of Cohn before operation, with repeated use of these procedures postoperatively. In addition, local hemostatic measures should be used.

It is obvious that if minor surgical procedures are accompanied by such risk of fatal hemorrhage, major operations must be proportionately more hazardous. The site of trauma is probably the most important criterion of operability. If the operative site is exposed so that local hemostasis, pressure, and application of hemostatic substances can be employed, the chances of staunching the flow of blood are vastly increased. Thus Firor and Woodhall[2] successfully carried out the amputation of the thumb in a hemophiliac; Davidson and Levenson[3] have succeeded in skin grafting a hemophiliac with the help of thrombin; and Blalock[4] in 1932 amputated the arm of a proven hemophiliac. The occurrence of hemorrhage internally, either spontaneously or secondary to trauma, where local controlling measures cannot be continually applied presents a much more serious and difficult problem of management. A review of the literature in an attempt to collect opinions and results of major surgery in cases of hemophilia yields conflicting data. It is apparent that a wide discrepancy exists in the minds of the various authors as to the efficacy of surgery in acute emergencies, varying from almost complete lack of restraint

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in regard to operability to a feeling that no operation should be considered under any circumstances. Because of lack of unanimity of opinion as to the best course to follow in hemophiliacs who develop acute surgical conditions, a thorough search of the literature has been made with the hope that definite conclusions might be reached.

It is unfortunate that in many instances sufficient evidence is not presented by the particular author to allow an unequivocal diagnosis of hemophilia to be made. In view of the necessity of accurate diagnosis in attempting to formulate an opinion as to the advisability of surgery in hemophilia, we have eliminated from the analysis those reports in which there is any doubt of the specific nature of the hemorrhagic diathesis. The criteria established for the diagnosis of hemophilia are clear cut. These will be discussed further together with the reasons for exclusion of questionable cases. Those reports in which the diagnosis of hemophilia was unquestionable and those cases in which operative intervention for some associated disease was carried out will be reviewed in some detail.

In 1931 Emile-Weil[5] presented the results of surgical treatment of various disorders in an extensive article. The most outstanding case was one in which a gastro-enterostomy was performed because of a bleeding prepyloric ulcer. The operation was successful, hemorrhage being controlled by transfusions. In this case the coagulation time was two hours; clot retraction was good; bleeding time was three to seven minutes. Family history and past history were dubious. It seems probable that this was a case of true hemophilia. Another case reported by the same author involved the operative removal of a gangrenous appendix with recovery of the patient. This patient was five years of age; the family history, past history, and the sex of the patient are not given. The clotting time was 45 minutes; the bleeding time, nine minutes. Insufficient data are given to establish the diagnosis beyond doubt in this instance, although the prolonged coagulation time is highly suggestive.

The case of Blalock[4] reported in 1931, and already mentioned, was undoubtedly one of true hemophilia. However, here the application of local measures was possible; and although the procedure was major, it is not comparable to one accompanied by internal hemorrhage.

Cioran[6] in 1935 reported a case of acute appendicitis in a soldier shown to have hemophilia after operation. Bleeding of more than the usual amount was noted during operation, and later it was found that the coagulation time was four hours. Platelet counts were normal. It was determined that a maternal grandfather had a questionable hemorrhagic history and had died at an early age. The patient's past history revealed that he had had repeated nose bleeds and bruised easily during childhood. The patient was given two transfusions postoperatively without effect on the hemorrhage. However, intramuscular and intravenous injections of a substance called "Clauden" controlled the bleeding. This substance, "Clauden," was said by the author to be thrombokinase (thromboplastin) and was an aqueous extract of lung tissue. The patient gradually convalesced over a 35-day period. There is little doubt that this case was one of true hemophilia. It is rather startling

that hemorrhage should have been controlled by intravenous injections of thromboplastin, a procedure which has been shown in animals to be accompanied by intravascular thrombosis and death if given in large amounts.

Friedrich[7] in 1935 published an extensive article on the risk of operation in hemophilia. He cited several cases of his own and reviewed the literature, and arrived at some very definite conclusions. However, he made no attempt to present data supporting the diagnosis of hemophilia in these cases. He estimated the overall mortality of all major surgical procedures in hemophiliacs at 35 per cent. He felt operation in cases of appendicitis contraindicated except in the event of peritonitis, and cited two appendectomies which terminated fatally. He felt that the risk of hemorrhage following surgical intervention in appendicitis was greater than the risk of infection with conservative treatment (and this at a time before antibiotics and sulfonamides were available for clinical use.) He also found reports of six cases of retroperitoneal hematomata which were incorrectly diagnosed as acute abdominal emergencies, and were operated upon with uniformly fatal results. His opinion was that abdominal surgery should not be undertaken unless it was evident that death would ensue if surgical measures were not taken to alter the course. He pointed out that only in those cases where local measures could be applied was the prognosis favorable following surgery.

Mertz and Meiks[8] in 1938 reported a hemophiliac in whom nephrectomy was necessary because of a severe left hydronephrosis with infection. The diagnosis in this case was adequately established, and treatment was thorough and according to the accepted principles. This report will be quoted in some detail because of the striking similarity between the course of their patient and that of the case included in this paper. The patient was carefully studied and prepared preoperatively by means of sensitization to sheep serum and transfusion. The clotting time was brought down from 120 minutes to five and a half minutes prior to operation. Especial care was taken by the surgeons to obtain complete hemostasis, and no unusual bleeding was apparent at operation. Postoperatively, the patient's clotting time was six minutes and was kept down to low levels by repeated transfusions. There were no signs of bleeding for three days, and the patient appeared to be recovering. The drain which had been placed at operation was removed. However, the patient began to run a fever as high as 104.4.° On the fourth postoperative day, it was thought that free abdominal fluid had collected. The signs of this became more definite later; the intra-abdominal hemorrhage progressed in spite of repeated transfusions, and the patient died on the twelfth postoperative day due to hemorrhage and streptococcal septicemia. Autopsy showed a large hematoma at the site of the removed kidney with no evidence of bleeding from a major vessel but merely a slow ooze from the renal bed. The authors emphasized the fact that bleeding continued despite repeated transfusions and a clotting time at the lower limits of normal.

Vance[9] in 1939 reported four cases of surgical conditions in male patients with abnormal bleeding. One underwent a radical mastectomy because of carcinoma. He experienced some bleeding postoperatively which gradually

stopped without the necessity of transfusion. The clotting time in this patient was 16 minutes; family history and past history negative. The second patient had acute appendicitis. The clotting time in this case was only six minutes (method not given) and yet the diagnosis of hemophilia was made. Postoperatively the patient did well for five days but then bled continuously into the abdomen, in spite of blood transfusions, and died on the ninth postoperative day. The third case was one of acute appendicitis with a history very suggestive of hemophilia and a coagulation time of 18 minutes. Family history showed that a brother also had a hemorrhagic tendency. He was treated conservatively, with appendiceal rupture and subsequent localized peritonitis. The patient apparently bled considerably per rectum in spite of repeated transfusions and continued to do so for two weeks. Following this he rapidly improved. The fourth case was a brother of the above patient who also developed appendicitis. He was treated in the same manner as the previous case and died at the end of eight days. It is probable that these last two patients with appendicitis were true hemophiliacs. In respect to the first two patients, however, the diagnosis, if made on the basis of the data presented, is less certain.

Birch[1] in her monograph mentions one case of acute appendicitis in a true hemophiliac. Although the details of this case and the operation are not given, apparently the patient recovered from the operation but died one week later from postoperative pneumonia with hemorrhage from the lungs. It is doubtful that this case can be justly classified as an operative success.

A German military surgeon, Karitzky,[10] reported the treatment and course of a hemophiliac who had received severe combat wounds in October, 1939. There could be no doubt of the accuracy of the diagnosis in this case, the past history, family history and laboratory findings being classical of hemophilia. This marine had been injured by the explosion of a mine under his ship, steel splinters entering the left neck and hip regions. The steel fragment entering the hip region had penetrated, leaving a large wound of exit in the gluteal region near the anus. Hemorrhage was intense, and the patient lapsed into shock. Débridement was carried out, the fragment removed from the neck with closure; and the gluteal wound was packed with iodoform gauze. The cervical wound healed normally. On the fifth day after injury, the patient began to lose large amounts of blood from the gluteal wound. He continued to bleed for 24 days and became dangerously anemic in spite of repeated transfusions and injections intramuscularly of substances such as Redoxon, Koagulen, Gelatine, Clauden, etc. A putrid wound infection developed. Massive transfusions and pressure dressings finally controlled the bleeding to a large degree, but oozing from stitch wounds and needle punctures continued for some time. The patient finally recovered. This example of protracted uncontrollable hemorrhage from a wound which was amenable to local measures in a hemophiliac treated intensively with blood transfusions again demonstrates the serious difficulties which may be encountered in bringing about hemostasis. However, the fact that no internal operative procedures were performed in this case removes it from that type of operative intervention with which we are concerned.

In addition to those reports reviewed, there have been numerous others of major surgery performed in patients suffering from some hemorrhagic diathesis. The nature of the surgical condition, the results of surgery, and the data upon which the diagnosis of hemophilia was made are presented in Tables 2 and 3. It was our opinion that insufficient information was presented by the authors for the diagnosis of true hemophilia in these instances. The criteria necessary for the diagnosis of this particular hemorrhagic diathesis and the reasons for excluding these cases will be discussed in more detail later.

PRESENTATION OF A CASE

E. W., a 27-year-old single white male, was admitted to Strong Memorial Hospital October 4, 1947. He had been awakened the previous morning by a crampy epigastric pain which was followed by nausea and vomiting. The pain persisted after he had vomited, was generalized throughout the abdomen but was slightly more pronounced in the right lower quadrant. He had taken a dose of cascara which he had promptly vomited. In spite of a small but normal bowel movement that day, his pain became steadily worse. On the morning of admission the pain was localized in the right lower quadrant, was constant and rather dull and aching in character. He had felt feverish but had no other complaints.

Past History: The patient had been seen in this hospital in 1927 at the age of seven with a history of having bled profusely and bruised easily since early childhood. The coagulation time of whole blood was found to be 4 hours, and a diagnosis of hemophilia was made at that time. He had had numerous hospital admissions and clinic visits from that time forth because of hemoarthrosis, hematuria, and hemorrhage from the gums. He was intensively studied in 1940 and again in 1945, and had received extensive orthopedic care. As a result of the use of fresh plasma and whole blood and of careful orthopedic management, he had been able to work fairly regularly and to walk during much of this time. He had been free of bleeding during these periods of therapy for as long as one year. Fresh whole blood or plasma always lowered the coagulation time. In spite of 6 whole blood transfusions and 100 plasma transfusions from 1927 to the present time, no refractory state ever developed. A detailed summary of the laboratory findings is found in Table I.

TABLE I

	1927	1939	1940	1941	1945
Coagulation time	40′	30′	70′	85′	36′–72′
Clot retraction	good	good	good	good	good
Bleeding time	3.25′	1.5′	....	....	....
Prothrombin concentration (% normal)	....	normal	....	....	80 per cent
Calcium	....	10 mg. per cent	....	....	10.4 mg. per cent
Fibrinogen	....	....	....	....	0.2 Gm. per cent
Vitamin C	....	....	....	....	.76 mg. per cent
Platelets	250,000	normal no.	normal no.	normal no.	normal no.

Family History: revealed that a maternal great-uncle had "bled to death," but no other important family history could be obtained. This is shown in Figure I.

Physical Examination: revealed a moderately well developed, somewhat dehydrated 27-year-old white male in moderate distress. There was a fine macular rash over the anterior chest, and there were ecchymotic areas over the left hand. His temperature was 101.3° F. The blood pressure and pulse were normal. The pertinent physical findings were limited to the abdomen and the extremities.

The abdomen was slightly distended, tender in the epigastrium and more so in the right lower quadrant where there was also spasm and rebound tenderness. There was bilateral rectal tenderness, more marked on the right. Both ankles and both knees showed old joint deformities.

The white blood count was 14,200 per mm.^{3} at the time of admission. The urine was negative except for a strong acetone reaction. In view of this patient's known hemophilia, it was decided to employ conservative measures even though it was felt that he had acute appendicitis. Accordingly he was given penicillin 100,000 units intramuscularly every three hours, was hydrated, and given 5 grams of sodium sulfadiazine in 1000 cc. of saline intravenously. It was decided to reduce the coagulation time, which was found to be 60 minutes. Four-tenths Gm. of Fraction I of Cohn ("Antihemophilic globulin") and 100 cc. of freshly prepared plasma were administered intravenously. The coagulation time was 11 minutes when observed 20 minutes after this treatment and was 15 minutes 4 hours later.

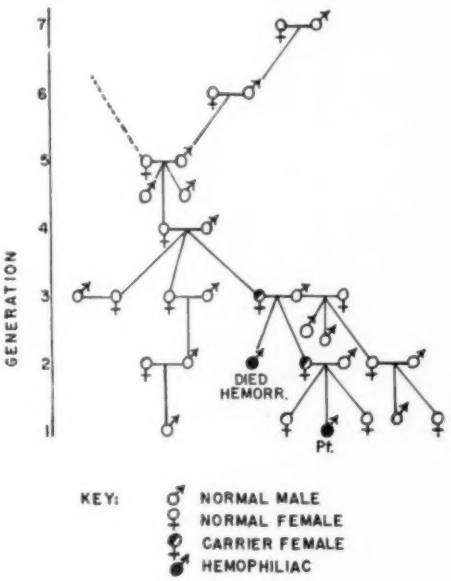

FIG. 1.—Graphic representation of patient's family history.

After a period of observation of 10 hours, during which the leukocytosis, the fever and the symptoms increased, it was decided to operate. This decision was reached after much deliberation and in view of the normal coagulation time, although it was felt that there was considerable risk involved in the procedure.

Operative Note. Under ether anesthesia, the abdomen was opened through a small McBurney incision. No unusual bleeding was encountered. The peritoneal cavity contained a small quantity of cloudy fluid which on culture was found to contain *E. Coli.* The cecum was delivered into the wound without difficulty. The appendix lay in a retrocecal position with its tip beneath the ileocecal valve and surrounded by fairly dense adhesions. These adhesions were broken up by blunt dissection, but the tip of the appendix could not be mobilized. A small Weir extension was made for better exposure, in spite of which the appendix could not be delivered. It was thought unwise to mobilize the cecum by incising its lateral attachments because of the danger of hemorrhage. For this reason the appendix was removed in a retrograde fashion partly by blunt dissection. Its wall was necrotic at the site of two fecoliths near its mid-portion. The stump was ligated with a surgeon's knot of 000 silk and inverted beneath two purse string sutures. There was a moderate ooze from the appendiceal bed which could not be visualized. It did not appear to be dangerous in quantity and was not felt to be arising from any sizable vessel. It was satisfactorily controlled with a pack of gelfoam. Drainage was discussed but not carried out. The abdominal wall was closed in layers with silk, taking great pains to ligate or cauterize the smallest bleeding points. In spite of this care, there was minimal ooze from the subcutaneous tissue.

The patient's condition was good at the close of the procedure. Pathologic examination of the specimen confirmed the clinical diagnosis of acute appendicitis.

During the operation the patient received 500 cc. of fresh compatible whole blood, and one hour later he was given 0.2 Gm. of Cohn Fraction I and another 500 cc. of fresh compatible whole blood. He was placed on Wangensteen suction. Careful check was kept of the coagulation time (the Lee-White method, performed at 37.5° C. and by the same person as often as possible) and the plasma and whole blood specific gravity. These, in addition to the antihemophilic therapy, are summarized in Figure 2. At no time could a circulating anticoagulant be demonstrated similar to that described by Craddock and Lawrence.[19]

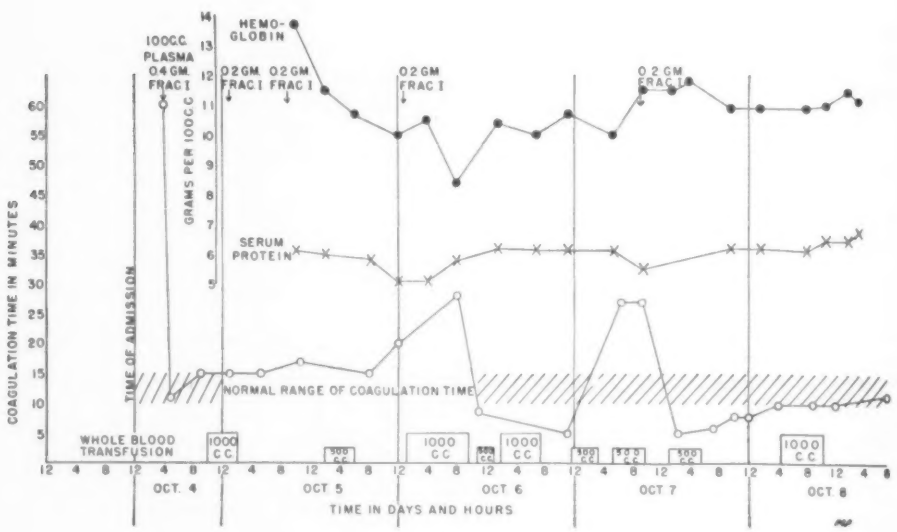

FIG. 2.—Course of patient in hospital showing amounts of antihemorrhagic treatment, response of coagulation time, and degree of serum protein, and hemoglobin deficiency.

Penicillin was continued, but because of the presence of albuminuria and microscopic hematuria, sulfadiazine was discontinued. A conscientious attempt was made to maintain a normal fluid and electrolyte balance.

In spite of the fact that the coagulation time was kept almost entirely within normal limits, the patient continued to lose blood into his peritoneal cavity and into his abdominal incision. His abdomen became distended to the point where he had severe respiratory embarrassment, and on the fourth postoperative day it became necessary to perform an abdominal paracentesis, 600 cc. of bloody, incoagulable fluid being removed. The patient experienced some relief, but later in the day he was again severely dyspneic; and there were signs of atelectasis. The abdominal wound was partially opened under sterile precautions and 600 cc. of fluid, defibrinated blood removed, this later proving to have been extraperitoneal.

On the evening of the fourth postoperative day, the patient's respiratory embarrassment became extreme; there were signs of bilateral pleural effusion, and thoracentesis was performed. After 150 cc. of bloody fluid had been withdrawn, the patient's respirations ceased. Artificial respiration and intravenous coramine were of no avail, and the patient expired.

Post mortem Examination: revealed pulmonary edema, bilateral hemothorax, a large blood clot in the appendiceal bed extending along the right peritoneal gutter from the pelvis to the liver. There was no evidence that the bleeding had come from any large bleeding point. There was also a tremendous hematoma at the site of the abdominal wound, not communicating with the peritoneal cavity.

DISCUSSION

In reviewing the literature on this subject, we have attempted to analyze critically each report in order to determine which cases represented operation in true hemophilia. The criteria selected by us for the establishment of the diagnosis of hemophilia in the individual cases reviewed are the following (see Table II.) (1) Laboratory findings showing definite prolongation of the clotting time as the only abnormal test of those routinely performed pertaining to the hemostatic mechanism. (2) Positive family history (although occurrence of sporadic spontaneous cases is occasionally found.) (3) A past history typical of that exhibited by the majority of hemophiliacs. (4) Occurrence in a male. These criteria are taken because the majority of the reports do not include detailed clinical and laboratory data in regard to the question of hemophilia. Attempt has been made to weigh the facts given in each instance and determine whether or not they permit the diagnosis of hemophilia.

It is recognized that there is great variation in the technic and results obtained in tests performed in regard to the clotting mechanism. Nevertheless, the one laboratory abnormality necessary for the diagnosis of hemophilia (in the untreated case) is a prolonged clotting time of the blood. In most instances the technic used for this test was not stated, but unless a definite prolongation over the stated limits considered normal by the author is recorded, the laboratory data cannot be regarded as diagnostic unless accompanied by other tests supporting the contention. In true hemophilia all the other tests routinely performed pertaining to coagulation time are normal (*i.e.*, prothrombin time, bleeding time, tourniquet test, platelet count, and clot retraction, and determinations of calcium and fibrinogen concentrations.)

In those cases reported, if the laboratory data are of questionable significance, the other criteria are studied. If there is a definite positive family history, that is, reliable evidence of hemorrhagic disease in male members of previous generations on the maternal side, then this is considered to support the diagnosis of hemophilia in the face of positive or suggestive laboratory data. Here again in many reports, the family history was either not mentioned or else was not elucidated in questionable instances. It is recognized that sporadic cases occur in whom no family history is detectable no matter how far back it is investigated. For this reason a negative family history does not exclude the diagnosis, but a positive one is good supportive evidence. In regard to the criterion of past history in the particular cases reviewed, it is felt that most hemophiliacs present stories of previous hemorrhagic phenomena which are fairly typical of the disease. Such occurrences in the past history as repeated hemarthrosis, subcutaneous bleeding after slight trauma, spontaneous bleeding per rectum or hematuria without the

presence of pathologic lesions, and prolonged bleeding from minor cuts are usually cited by the hemophiliac. The absence of such a past history by no means excludes the diagnosis, but its presence lends support in questionable cases. However, such symptoms can occur in other hemorrhagic diseases, and for this reason a positive past history alone cannot be used as the sole criterion for the diagnosis. The sex of the patient was not given in all reports. As far as is known at present, hemophilia occurs only in males. This has not always been accepted as true in the past. Therefore, in the older reports unless the sex is definitely stated or inferred, doubt is cast on the validity of the diagnosis of hemophilia.

ANALYSIS OF REPORTED CASES OF MAJOR SURGERY IN HEMOPHILIA

As shown in Table II extensive surgical procedures have been successful in areas where local measures could be applied to control blood loss. However, in regard to abdominal surgery, the chances for a satisfactory outcome are much less. If one accepts all cases reported as being true hemophiliacs, it would appear that the risks of major operation are not more extreme than would be indicated in the past.

Thus, of 15 major operations performed in areas of the anatomy where the application of pressure and local hemostatic agents was not possible, there were 11 recoveries and four deaths, or a mortality of 26.7 per cent. This mortality is close to that estimated by Friedrich for operations of this type in hemophilia. The percentage of recoveries is higher than might be anticipated. Indeed, from analysis of the cases, excluding those not proven to be true hemophilia, it is evident that the percentage is less optimistic. Of the 15 cases reported, there are only four which, on the basis of the facts presented, fulfill the criteria needed for the unequivocal diagnosis of hemophilia. Of these four, there were two fatalities which gives a mortality of 50 per cent. If our case is included, the mortality is raised to 60 per cent. All of these fatal cases received adequate treatment with transfusions. It would seem, therefore, that major surgery in cases of hemophilia of the type where local hemostatic measures cannot be utilized is accompanied by an expected mortality rate which is much greater than that which is usually indicated by the literature on this subject.

In regard to treatment of acute appendicitis *per se* in cases of hemophilia, the literature reviewed by us discloses 11 reports. Eight of these were operated upon and three treated conservatively. Of the eight receiving operative treatment, there were six recoveries and two deaths, or a mortality of 15 per cent. However, among the eight undergoing appendectomy, there were only two who fulfilled the criteria needed for the unequivocal diagnosis of hemophilia. Of these two, one died, and one recovered, thus giving a mortality of 50 per cent following operation. Our case would raise the mortality to 66.6 per cent.

On the other hand, there were three cases of true hemophilia with appendicitis treated conservatively. Two of these were definite instances of acute

TABLE II.—*Reported Cases of Major Surgery in Hemophilia.*

Associated Disease	Authors	Operation	Outcome	Criteria of Diagnosis of Hemophilia: Laboratory Data	Family History	Past History	Sex	Positive Diagnosis Hemophilia	
Gangrene, arm	Blalock[4]	Amputation	Recovery	+	+	+	♂	Yes	
Trochanteric hematoma	Emile-Weil,[11] Scemama	Incision and drainage	Recovery	+	+	+	♂	Yes	External operative site; local hemostatic measures applicable
Carcinoma of breast	Vance[9]	Radical resection	Recovery	±	O	±	♂		
War wounds	Karitzky[10]	Debridement Removal of foreign body	Recovery	+	+	+	♂	Yes	
Tuberculous knee	Friedrich[7]	Exploration of knee	Recovery	O	+	O	♂		
Bladder polypi, prostatic median bar	Mathe and Mitchell[12]	Polypectomy Fulguration of median bar	Recovery	−	−	±	♂		
Prostatic hypertrophy	Hinman[13]	Perineal prostatectomy	Recovery	−	±	+	♂		
Hydronephrosis with infection	Mertz, Meiks[8]	Nephrectomy	Death	+	+	+	♂	Yes	
Peptic ulcer with hemorrhage	Emile-Weil[3]	Gastro-enterostomy	Recovery	+	±	+	♂	Yes	
Pre-pyloric ulcer with hemorrhage	Wosnessensky[14]	Gastric resection	Death	O	O	O	♂		Internal operative site; local hemostatic measures not applicable
Appendicitis	Emile-Weil[3]	Appendectomy	Recovery	+	O	O	O		
Tonsilitis and appendicitis	Hays[15]	Tonsillectomy Adenoidectomy Appendectomy	Recovery	−	O	O	♂		
Appendicitis	Vance[9]	Appendectomy	Death	±	O	O	♂		
Appendicitis	Birch[1]	Appendectomy	Death*	+	+	+	♂	Yes	
Appendicitis	Prima[16]	Appendectomy	Recovery	O	O	O	♂		
Appendicitis	Cioran[6]	Appendectomy	Recovery	+	+	−	♂	Yes	
Appendicitis	Hipsley[17]	Appendectomy	Recovery	O	±	+	♂		
Appendicitis	Paz[18]	Appendectomy	Recovery	O	−	+	♂		
Ruptured spleen	Friedrich[7]	Splenectomy	Recovery	O	+	O	♂		
Gastro-intestinal bleeding	Friedrich[7]	Gastric resection	Recovery	O	+	+	♂		
Appendicitis	This report	Appendectomy	Death	+	+	+	♂	Yes	

* Patient survived operation one week, but died with postoperative hemorrhagic pneumonia.

Symbols: + = Positive findings. − = Negative Findings. ± = Questionable or inadequate findings. O = Data not given.

appendicitis. The other patient, that of Platou[20] was felt at first to have appendicitis with intestinal obstruction. The latter disappeared after Wangensteen suction was instituted, and the patient recovered. The authors later felt that this patient probably did not have acute appendicitis but rather a hemorrhage into the submucosal area of the small intestine. Therefore, the total number of cases of appendicitis in hemophiliacs reviewed by us who received nonoperative treatment is two, with one death.

TABLE III.—*Results of Treatment of Appendicitis in Hemophilia.*

Author	Type of Treatment	Outcome	Criteria of Diagnosis of Hemophilia: Laboratory Data	Family History	Past History	Sex	Positive Diagnosis Hemophilia
Hipsley[17]	Appendectomy	Recovery	O	±	+	♂	
Emile-Weil[5]	Appendectomy	Recovery	+	O	O	O	
Hays[15]	Appendectomy	Recovery	−	O	O	♂	
Vance[9]	Appendectomy	Death	±	O	O	♂	
Cioran[6]	Appendectomy	Recovery	+	+	−	♂	Yes
Birch[1]	Appendectomy	Death*	+	+	+	♂	Yes
Prima[16]	Appendectomy	Recovery	O	O	O	♂	
Paz[13]	Appendectomy	Recovery	O	−	+	♂	
This report	Appendectomy	Death	+	+	+	♂	Yes
Vance[9]	Non-operative	Recovery	±	+	+	♂	Yes
Vance[9]	Non-operative	Death	±	+	O	♂	Yes
Platou[20]	Non-operative	Recovery**	+	+	+	♂	Yes

* Patient survived operation one week but died with postoperative hemorrhagic pneumonia.
** The diagnosis of appendicitis was not felt to be definitely established by the author.
Symbols: + = Positive findings.
− = Negative findings.
± = Questionable or inadequate findings.
O = Data not given.

The small number of reported cases of acute appendicitis in hemophiliacs treated by either operative or nonoperative means does not allow any valid comparison of expected results. Nevertheless, it is obvious that the dangers of uncontrollable fatal hemorrhage following internal operative intervention multiply many times the operative risk. Most statistics on the subject place the present mortality rate following acute appendicitis with rupture and peritonitis at 10–15 per cent in otherwise normal individuals.[21-24] It is not known how much the presence of hemophilia would increase the dangers of peritonitis of appendiceal origin. Certainly there is no good evidence to suggest that hemophiliacs cannot handle infectious processes as well as normal individuals. From our review of the literature, we believe the mortality rate following appendectomy in hemophiliacs is around 50 to 60 per cent. It is probable that the chances of death due to postoperative hemorrhage outweigh the expected mortality associated with the nonoperative treatment of appendiceal rupture and peritonitis.

The hemophiliac presented by us had been followed in this hospital since the onset of his hemorrhagic difficulties and had been the subject of extensive investigative and therapeutic procedures. It was well known that he obtained marked benefit from plasma injections, and he had been kept entirely free of

hemorrhagic phenomena for long periods of time without ever showing evidence of becoming refractory. As shown in the case report, when he was given blood, his clotting time fell rapidly to normal. He was of the optimal age group for surgery and possessed the physical stamina to withstand such a procedure. It was our hope that if operation were necessary, the coagulation mechanism could be augmented by transfusion or injections of fresh plasma or Fraction I of Cohn to such an extent that severe hemorrhage could be controlled.

The persistence of hemorrhage in this patient in spite of a normal coagulation time demands further comment and brings to mind several considerations in regard to the present concept of the fundamental defect in hemophilia. It is the belief of many investigators in this field at the present time that the underlying abnormality in hemophilic blood is a deficiency or lack of a plasma factor necessary for coagulation to take place in the normal span of time. This factor has been shown, by numerous workers,[25-27] to be present in normal blood but deficient in hemophilic blood. Howell[27] worked extensively with this factor which he called "plasma thromboplastin." Quick[28] feels, as did Howell, that this factor represents an inactive form of thromboplastin which is normally present in the blood and is activated by an enzyme released from platelets when coagulation begins. The factor has more recently become available in purer form and in greater amounts through the fractionation of plasma by Cohn and co-workers at Harvard. The active principle is contained in greatest concentration in Fraction I and Fraction III, subfraction 2 of Cohn's classification. It has been termed "antihemophilic globulin" by Lewis, Tagnon, *et al.*[29] and has been investigated extensively by these and other workers.

There is no question that "antihemophilic globulin" in very small amounts has a beneficial effect on the clotting mechanism of hemophiliacs. This effect is demonstrated by the sharp fall in coagulation time to normal values within a very short time after it has been injected intravenously. However, its effect is no greater and of no more permanency than the beneficial response elicited by whole blood or plasma. This is as might be anticipated since each type of treatment merely supplies a factor which is deficient in hemophilic blood. This factor, necessary for coagulation to occur in the normal span of time, is apparently used up over a period of 12 to 48 hours, so that at the end of this time, the deficiency again exists; and the coagulation time again becomes prolonged.

The fact that the deficiency of "antihemophilic globulin" was erased by the concerted treatment given this patient is borne out by the shortening of the coagulation times to normal levels. The continuation of hemorrhage, even in the face of the normal coagulation time of the blood, forces one to the conclusion that the mere deficiency of the substance "antihemophilic globulin" cannot be the sole abnormality of coagulation in hemophilia. There are other, perhaps more subtle changes from normal in the coagulation mechanism which cannot be measured by the tests of blood coagulability routinely used. It is well to remember, therefore, that the actual time consumed by clot formation in the test tube is by no means an accurate measure of the ability of the hemostatic mechanisms to control blood loss.

Other recent investigations into the clotting mechanism have made it highly suggestive that the present concept is incomplete. Attempt will not be made to review these advances, but it is becoming apparent that there are factors concerned in the first stage of clotting that heretofore have not been appreciated. Thus Ware *et al.*[30] believe an activator globulin (AC globulin) is necessary for prothrombin activation; Milstone[31] postulates a prothrombokinase which is activated in the presence of calcium ions to thrombokinase (thromboplastin) and which in turn activates prothrombin; Owren[32] reports a principle designated as Factor V which is essential for rapid prothrombin conversion to thrombin, and Quick[33] has designated two components of prothrombin (A and B) as well as a "labile factor" which is also necessary for coagulation to occur in the normal time. It is probable that these various factors are closely related, if not the same. It is not known where these factors fit into the present formula of clotting; there is little doubt that the speed of prothrombin conversion and hence the speed of clotting is greatly influenced by their presence. These investigations as well as others such as those of Ferguson[34] indicate the existing confusion and inadequacy of knowledge concerning blood coagulation. The importance of these advances in regard to the treatment of hemophilia are unknown at present. Certainly this case would indicate that a mere deficiency of "antihemophilic globulin" does not explain the blood loss in all cases. It is likely that the normal fresh blood given to this patient in large quantities contained an adequate amount of all the factors described by various authors as necessary for coagulation. It is evident that whatever the defect may be which causes hemorrhage in hemophilia, it cannot always be corrected, even temporarily, by supplying the patient with those factors concerned with clotting as they exist in normal blood.

The main points to be emphasized with regard to this case are the following: First, and most important, the coagulation time of the blood cannot be taken as a measure of the adequacy of the hemostatic mechanism in the hemophilic patient. Howell[27] has stated, "at present the one pathologic condition in hemophilia that is definitely established and that is directly connected with the hemorrhage is the prolonged clotting time of blood. Those who have had the widest experience with hemophilic patients believe the severity of the disease is in general proportional to the delay in coagulation." This feeling has been almost generally accepted in regard to the disease. The main object of all therapy up to the present time has been to lower the coagulation time of the blood. This patient demonstrates clearly that coagulation time alone can be used neither as the sole measure of the severity of the hemorrhagic tendency nor as a test of the response of the hemophiliac to treatment. There can be no doubt that the coagulation time of this patient's blood was usually maintained within limits which are considered normal, and yet hemorrhage continued with loss of the blood as rapidly as it was replaced.

The second point to be emphasized is the difficulty in choosing a case for operation. By all the accepted standards this hemophiliac met most of the requirements for operability, as well as any patient suffering from this disease

could meet them. And yet death resulted in spite of most intensive antihemorrhagic treatment.

The third point to be stressed is the infinite difference between controlling postoperative hemorrhage from areas that are exposed and amenable to local measures and controlling hemorrhage from internal areas. It is our feeling that if the site of bleeding cannot be treated with pressure, application of hemostatic substances, and other local measures, then the chances of recovery from surgery are poor.

In view of the unfortunate results of surgery in this case and the unconvincing reports of the efficacy of major surgery in hemophiliacs as gathered from the literature, it is our belief that conservative medical treatment is the course of choice in these cases unless it becomes clearly evident that death will ensue if surgery is not undertaken. The dangers of uncontrollable hemorrhage following surgery outweigh the dangers of generalized peritonitis and subsequent fatal infection, especially with the modes of controlling infection now at hand. The only existing well established means of combating hemorrhage in these cases is to give the patient fresh whole blood transfusions, injections of fresh or lyophile plasma, or the substance known as "antihemophilic globulin" as contained in Fraction I of Cohn. Each of these three forms of therapy accomplishes the same end. Each supplies the hemophiliac with a deficient substance necessary for coagulation to take place in the normal span of time. When the hemophiliac is given this substance by any of the means described, the coagulation time of the blood is reduced to normal. And yet, as demonstrated by this case, correction of this deficiency with a concomitant reduction in the clotting time to normal does not always promote hemostasis. This lack of response on the part of the hemophiliac's clotting mechanism to present modes of therapy in all instances further increases the dangers associated with any type of major surgical procedure undertaken in patients with this disease.

SUMMARY AND CONCLUSIONS

1. A review of the available medical literature reveals that the reported instances of internal operative procedures in patients with hemophilia require careful analysis. Many of the cases are accompanied by data that are inadequate for the diagnosis of this hemorrhagic diathesis.

2. The mortality following internal surgery in established cases of hemophilia is relatively high. Of four previously reported cases in whom the diagnosis of hemophilia was unequivocal, two died from hemorrhage following operation while two recovered. The other eleven reports of internal surgical procedures in patients with bleeding tendencies were excluded from this analysis because of inadequate basis for the diagnosis of hemophilia. If these were included, the total number of recoveries following operation would be eleven with only four deaths, or a mortality rate of 26.7 per cent, a figure we believe to be erroneously low.

3. A case of unequivocal hemophilia is presented with the complication of acute appendicitis. The patient received intensive antihemorrhagic therapy,

but despite a normal clotting time he continued to bleed profusely and expired four days postoperatively.

4. A discussion of the significance of continued hemorrhage in the presence of a normal *in vitro* clotting time and of its relation to the fundamental defect in hemophilia is presented. Emphasis is placed on the failure of the coagulation time to indicate the severity of the hemorrhagic tendency or the degree of response to treatment, the difficulty in choosing a suitable case for operation, and the great difference in controlling internal hemorrhage as opposed to bleeding from an external site.

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REDUCTION OF INTUSSUSCEPTION BY BARIUM ENEMA*

A CLINICAL AND EXPERIMENTAL STUDY

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IN 1836 THERE APPEARED in the American Journal of the Medical Sciences an account[1] of "A Case of Introsusception in Which an Operation Was Successfully Resorted to by John R. W. Wilson, M.D., of Rutherford County, Tennessee in December, 1831." The author concluded, "His recovery was rapid and entire. . . . The success of this case in which the operation was so long deferred, and at last performed under such unfavorable circumstances, warrants the propriety of resorting to it in the disease, and proves that relief may occasionally be afforded by this means when all others have failed."

Seventy-five years later, in 1906, operation was still so uncertain and dangerous that Harvey Cushing, as a house officer, wrote a note in the history of a child with intussusception, stating that operation was postponed with the hope that sphacelation might take place and the sphacelus be passed entire per rectum. As far back as 1784 Nuck[2] had diagnosed intussusception in a man of 50 years who was successfully operated upon by an unnamed surgeon. In 1825 Fuchsius[3] operated successfully for intussusception in a man of 68. Wilson's is the third successful instance in the literature and the first in this country. A few other operative cases, all fatal, had been reported by 1871 when Jonathan Hutchinson[4] performed the first successful operation for intussusception in a child, a baby of two years. Until this time the disease had been treated either expectantly, waiting for gangrene or sphacelation of the intussusception, or by rectal instillations. With this background Hirschsprung[5] of Copenhagen in 1876 reported his experiences with the treatment of intussusception by hydrostatic pressure. This method is older than any operative one. Hippocrates advised forced injection of water or air into the intestines in all forms of ileus, and several authors in the early nineteenth century advised and used clysters successfully. Others used bellows, or connected hydrogen sulfide generators with the rectum, or inserted effervescent powders. Still others passed long bougies to perform a direct reduction, or stimulated the intussusceptum with electricity, or tried to shrink it with cold water or hypertonic saline solution. By 1905[6] Hirschsprung was able to report on 107 personal cases of intussusception. His results were so superior to those previously reported that his contemporaries doubted his conclusions. He was forced, in self-defense, to publish a table listing each of his 107 cases. He presented a 35 per cent mortality in a disease which up to that time was fatal in almost 90 per cent of the cases. In 84 patients treated by Hirschsprung with

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enema alone, the mortality was 23 per cent. These figures compare favorably with those from the Johns Hopkins Hospital in the period from 1929 to 1938, more than 25 years after Hirschsprung.

Our interest in intussusception was aroused in 1933 by the paper of Dr. E. H. Miller[7] of Chicago who described his own poor results (45 per cent mortality in 20 cases) and referred to the remarkable results of Hipsley in Australia with reduction of intussusception by hydrostatic pressure. The method of reduction by hydrostatic pressure was systematized by the Danish school, Hirschsprung[5] and his successors, Kock and Oerum,[8] and Monrad.[9] All of these also practiced, to varying degrees, manual disinvagination of the intussusception through the intact abdominal wall. In 1913 the first report of intussusception diagnosed by roentgenography was made by Lehmann.[10] In 1927 Pallin and Olsson[11] in Sweden, Retan[12] in the United States, and Pouliquier[13] in France all reported reductions by barium enema under fluoroscopic control. The Australians continue to use simple saline enemas, but the use of the barium enema has gained great popularity in Scandinavia and South America. In this country, in England, and in Germany the use of hydrostatic pressure has never been accepted. It is a remarkable fact that surgeons, reporting in 1930 and 1940 a 20 and 30 per cent mortality for the treatment of intussusception, scorn the non-operative methods which had a lower mortality in 1890 and 1900 and which, in 1926, gave a mortality of as low as 5 per cent in 100 consecutive cases treated by Hipsley.[14] Hipsley, be it noted, took up the use of hydrostatic pressure to improve his mortality of 8 per cent with operative treatment. One of Hipsley's papers, published in 1935,[15] is of particular interest and should satisfy those who maintain with Ladd[16] that the situation is different in Australia because there an entire generation of medical men has been intensely interested in intussusception through the work and teaching of Sir Charles Clubbe.[17] Hipsley reported on this occasion 486 cases of intussusception treated by 10 surgeons with a mortality of 11 per cent. Three of the 10 surgeons always attempted a preliminary reduction by saline enema and had a mortality of 9.8 per cent. The other seven surgeons operated on all patients without injection and had a 14 per cent mortality. This is a significant difference in a large series of patients treated in the same period of time at the same hospital.

Table I tabulates the mortality from 10 American and British clinics, with figures running from 60 per cent to 8.3 per cent. The lower figures are all in recent years, but mortalities of 40 and 50 per cent are common 20 and 30 years after Hirschsprung's day. Table II, by contrast, shows the mortality in those clinics using hydrostatic pressure. There is a striking difference, the range being much lower with mortalities years ago of 5 per cent, 7 per cent, and 9 per cent. Most of the fatal cases were also operated upon; accordingly, it is to be expected that with modern operative and supportive methods even lower mortalities can be expected. It must also be remembered that mortality rates alone do not tell the story. The morbidity is many times higher in the operative cases—wound infections, distention, fever, postoperative intestinal obstructions (sometimes several bouts in the same child over a period of years).

The hospital stay of the operative patient is much longer. The average hospital stay in our 27 patients treated primarily by enema was 9.2 days, as opposed to an average hospital stay of 32.4 days for 16 others treated during the same period primarily by operation.

TABLE I. *Mortality from intussusception treated primarily by operation. Note the relatively high mortality rates compared with those in Table II.*

Author	Primary Method	Period	Number of Cases	Per Cent Mortality	Source
Kahle	Operation	1904–1938	150	52	New Orleans
		1928–1938	59	30.5	(Charity)
Perrin & Lindsay	Operation	1903–1920	400	34.8	London (London Hospital)
		1915–1920	113	22.0	London
Close	Operation	1904–1927	363	31	(Guy's Hospital)
Ladd & Gross	Operation	1908–1939	484	?	Boston
		1908–1912	?	59	(Children's Hospital)
		1928–1932	?	14	
		1939	12	0	
Peterson & Carter	Operation	1908–1932	64	30	New York
		1922–1932	21	10	
Wakeley & Atkinson	Operation	1920–1929	121	10	London (King's College)
Gordon	Operation	1917–1938	44	23	New Haven (Yale)
Miller	Operation	1934	20	45	Chicago
Gibbs	Operation	1927–1942	91	30	Cincinnati General
		1937–1942	36	8.3	
Mayo & Woodruff	Operation	1941	55	23.6	Mayo Clinic

TABLE II.—*Mortality from intussusception treated primarily by hydrostatic pressure. Note the low mortality rates and the early dates at which these were achieved.*

Author	Primary Method	Period	Number of Cases	Per Cent Mortality	Source
Hirschsprung	Saline enema Manual disinvagination	1871–1905	100	35	Copenhagen
Clubbe	Enema or operation	1901–1926	834	14.5	Australia
Hipsley	Saline enema	1926	100	5	Australia (Sydney)
Hipsley	Saline enema	1921–1934	171	9.8	
	Operation	1921–1934	315	14	Australia (Sydney)
Sjöström	Barium enema	1927–1934	38	16	Sweden (Lund)
Hellmer	Barium enema	1933–1942	110	7.3	Sweden (Lund)
Nyborg	Barium enema	1930–1941	108	7	Sweden (Lund)
	Barium enema	1935–1941	84	5	

Table III records the mortality from intussusception at the Johns Hopkins Hospital since its opening. From 1893 to 1928, long after Hirschsprung's death and after Clubbe's report from Australia of 834 cases seen over 25 years with a mortality of 14.5 per cent, a line of surgeons at the Johns Hopkins Hospital, including Halsted, Cushing, Follis, Heuer and others, had a mortality of 44 per cent. The next group of surgeons in the period 1929 to 1938 lost 14 out of 44 patients, a mortality of 32 per cent, and the last group, 1939 to 1947, lost 5 out of 48, a mortality of 10.4 per cent. Of these 48 patients, however, 27 were treated primarily by enema with no deaths, and 21 primarily by operation with five deaths or a 24 per cent mortality. It is only fair to note that

there has been some selection of cases in these two groups. Beginning in 1939 the senior author has administered a barium enema to every patient with intussusception whom he has seen. However, some residents chose to do primary operations on the patients they judged to be the sickest, whereas others operated on all patients with intussusception.

The barium technic is as follows: As soon as the presumptive diagnosis of intussusception is made, the child is taken to the fluoroscopic room. At the same time, if it is at night, the operating room is alerted. In the fluoroscopic room, without anesthesia, an unlubricated Foley bag catheter is inserted in the rectum. The bag is distended with 20 to 40 cc. of air according to the size of the patient, and barium is permitted to run in from a height of three feet. (Fig. 1.) It is important not to lubricate the catheter as it will then be more easily retained. In addition, it is essential for an assistant to squeeze the buttocks together throughout the procedure. The barium usually runs rapidly

TABLE III.

THE JOHNS HOPKINS HOSPITAL

MORTALITY FROM INTUSSUSCEPTION

1893 - 1928
24/54 PATIENTS DIED - MORTALITY 44%

1929 - 1938
14/44 PATIENTS DIED - MORTALITY 32%

1939 - 1947
21 OPERATIVE REDUCTIONS
5 DIED - MORTALITY 24%
27 ENEMA REDUCTIONS - MORTALITY 0

1939 - 1947
COMBINED MORTALITY 10%

to the intussusception, which it outlines, producing a concave meniscus. The ends of the meniscus extend proximally until suddenly the intussusceptum gives ground. The meniscus flattens out, then begins to deepen again, and so on. This may take place very quickly. An intussusception protruding from the anus was on one occasion reduced almost too quickly to be followed fluoroscopically. At other times there is a stubborn hang at one level or another and then sudden progress. At times a bubble of air precedes the barium column and alters the fluoroscopic picture. The chief difficulty may be in deciding upon completeness of reduction at the cecum. By displacing the cecum laterally one may see barium well up in the small bowel, and in one of our youngest infants barium went up into the proximal jejunum. If complete reduction is not achieved, the child is permitted to expel the enema and the

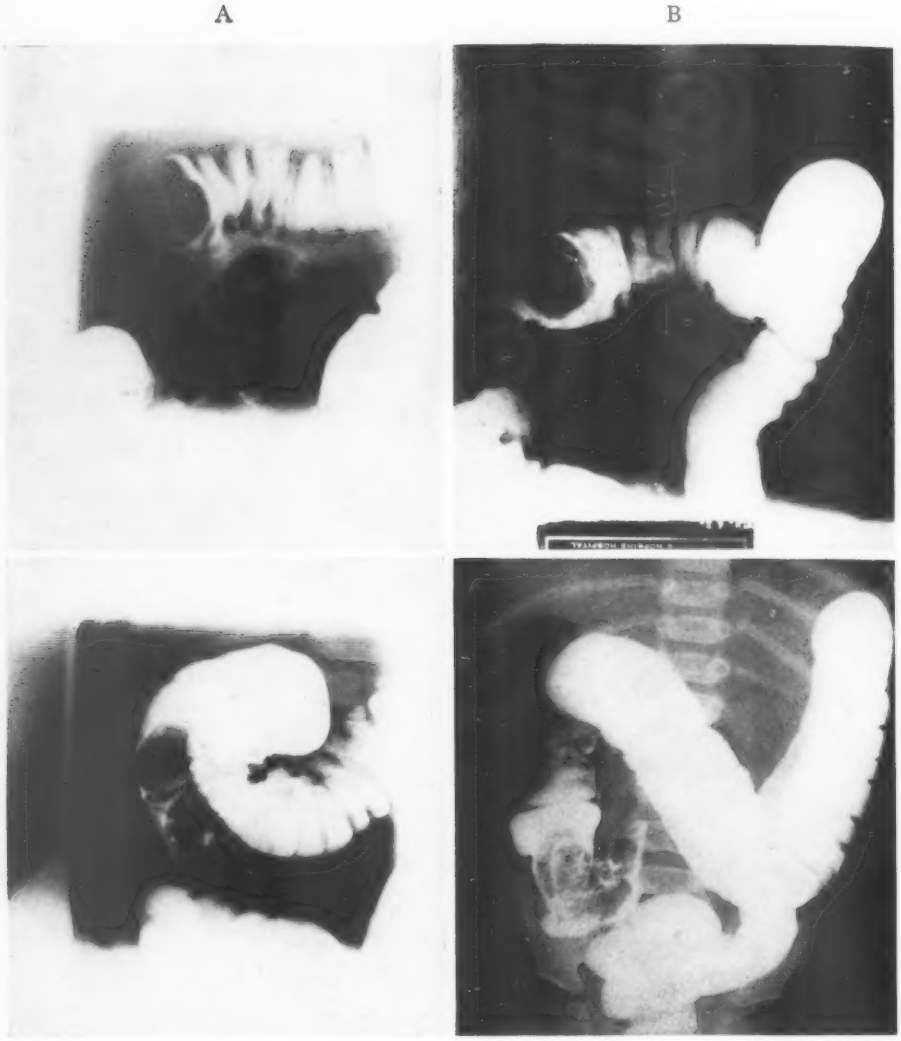

FIG. 1.—Barium enema of an intussusception which has reached the transverse colon.

A. The barium column meets the intussusception and the head of the column shows a concave meniscus.

B. The intussusceptum has been pushed to the hepatic flexure. The barium meniscus extends alongside the intussusceptum.

C. The reduction has proceeded to the ascending colon.

D. The cecum fills irregularly. A mass was still palpable. Through a McBurney incision an annular segment of ileum was pushed back through the ileocecal valve.

effort is repeated. If the third such injection is unsuccessful, operation is performed. After reduction the child is sent to the ward, powdered charcoal is instilled in the stomach and the administration of sulfasuxidine or oral streptomycin is begun as a prophylaxis against enteritis. Six hours later an enema is given to recover the charcoal. A successful reduction is denoted by the following criteria: (1) The entrance of barium well into the small bowel. (2) At times the child returns the barium with feces or flatus, proof in acute intussusceptions that the obstruction has been relieved. (3) The mass is no longer palpable. (4) The child is clinically relieved, often falling off to sleep.

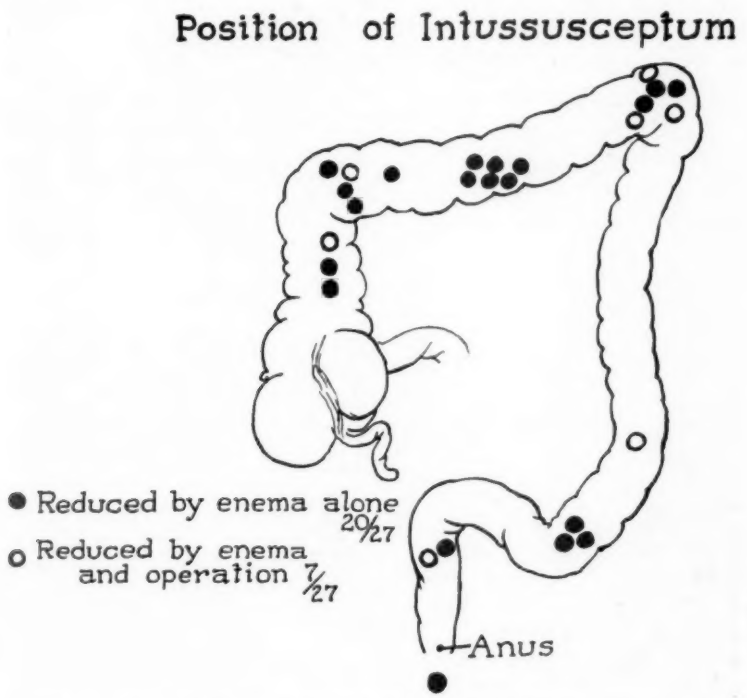

FIG. 2.—In three of the successful reductions by barium enema alone the intussusceptum had reached the sigmoid, in one it had reached the rectum, and in another had prolapsed through the anus. All of those patients requiring operative completion of the reduction were found to have had their intussusception reduced to the cecum or ileocecal valve.

(5) The subsequent recovery in the stool of charcoal given by mouth or the appearance of a blood-free stool is proof of reduction. Hipsley does not use fluoroscopy at all but depends upon the other criteria plus an additional one, namely, if sufficient fluid is injected, that which passes through the ileocecal valve will not be expelled for some time so that there is, after reduction, a persistent measurable increase in abdominal girth.

In the present series 27 intussusceptions were treated primarily by barium enema. All 27 patients survived. Twenty had complete reduction by enema—79 per cent, which is somewhat higher than the figure usually reported. The

diagram (Fig. 2) shows the level which the apex of the intussusceptum had reached when reduction was attempted. It will be seen that complete reduction was secured in a number of patients whose intussusceptions were at the anus, rectum, or sigmoid. Five of the 20 patients whose intussusceptions were reduced by enema alone, were of special interest. In two we were not certain reduction was complete and a McBurney incision was resorted to for verification of the reduction. In one patient, as an experimental procedure, the

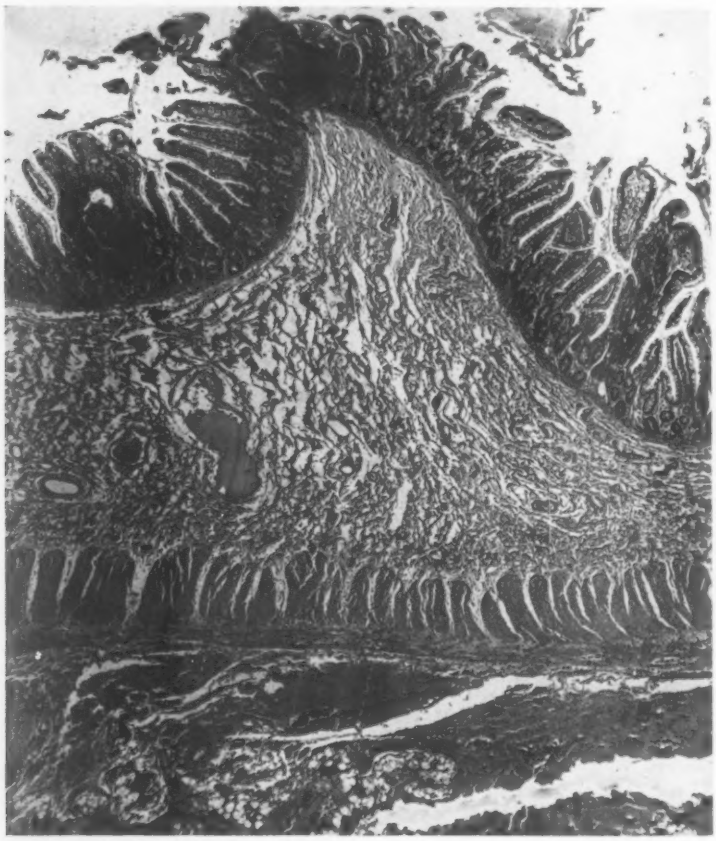

FIG. 3.—Photomicrograph x30 of bowel resected two days after barium enema reduction of intussusception. Child had been having bloody stools after reduction. Thickened bowel was found, thought to be involved in a neoplastic process, and resected. Section shows some sloughing of the tips of villi, massive submucosal edema, subserosal edema, and hemorrhage. Similar changes are regularly seen in animals after reduction of viable bowel.

abdomen was opened, the intussusception in the descending colon brought into view, and then a saline enema administered, reducing the intussusception under direct vision and without manipulation. We were astonished at the swiftness and gentleness of the reduction. A fourth patient had a recurrence two days later, after being symptom free and passing normal stools and the ingested charcoal. Another enema now reduced the intussusception to the cecum, and

exploration showed the appendix still intussuscepted. The appendix was removed and the patient recovered uneventfully. The fifth patient was operated upon 48 hours after enema reduction because of persistent pain, bloody stools, and a palpable right colon. We did not then know that these symptoms are a not rare consequence of reduction by any method. The operator resected the terminal ileum and right colon for great thickening, thought

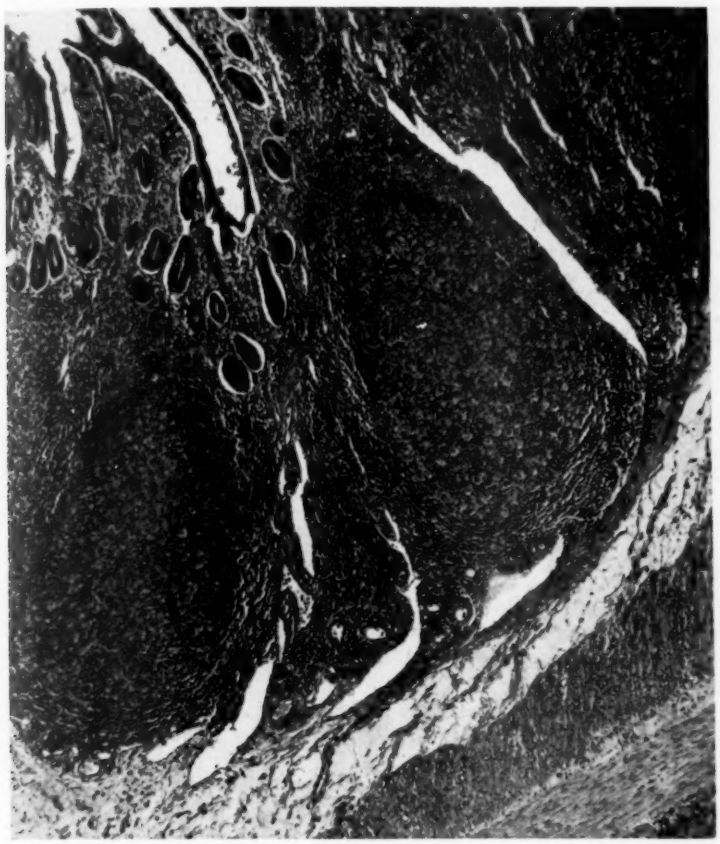

FIG. 4.—Photomicrograph x60. Section from annular segment of bowel removed at time of operative completion of reduction. The bowel shows early changes of intussusception—loss of epithelium, lymphoid hyperplasia, round cell infiltration in mucosa, submucosal and serosal edema. The bowel was viable.

to be due to tumor. As the photomicrograph (Fig. 3) shows, this was only the edema and organizing hemorrhage which follow reduction. We have reported elsewhere[18] changes much more extreme in the viable bowel of dogs several days after reduction of intussusceptions. It is worth remarking in connection with this patient that diarrhea and even dysentery are not rare occurrences after any type of reduction. Eighteen per cent of our total number of patients had diarrhea after reduction and of six patients who had stool cultures, three had pathogenic organisms.

The seven patients who required operation to complete reduction also survived. In every instance it was found at operation that the intussusception had been reduced to the cecum or ileocecal valve, in five patients from the left colon and in two from the right (Fig. 2). In several instances the operator used a McBurney incision. In only one of these patients had we made a mistaken diagnosis of complete reduction. In him, after a loss of six hours, we then repeated the reduction to the cecum and operation was performed, completing the reduction of an inch of ileum. There was a difference of opinion at the operating table as to whether resection should be performed. The operator did resect an annular segment, section of which (Fig. 4) shows the changes seen early in intussusception when the bowel is viable.[18]

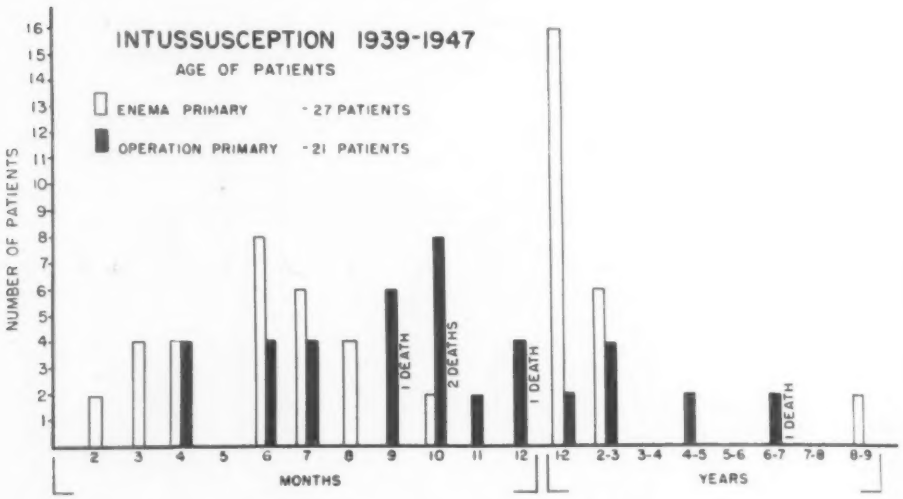

FIG. 5.—There is no marked difference in age distribution of patients in the two groups.

Figure 5 shows the distribution according to age of the patients treated by the two methods. There is no significant difference. Figure 6 shows the distribution according to duration of symptoms of the patients treated by the two methods. The group treated primarily by enema had more of the patients with symptoms of short duration. However, there were four patients in this group with symptoms of over 48 hours' duration.

The point of chief interest in the primary operative group of 21 patients is the history of the five fatal cases. In three the patient was sent home from this hospital the first time he was seen after the onset of symptoms of intussusception. In each instance the child was admitted the next day. Two patients were then operated upon promptly, but with the third there was a delay of an additional 47 hours. A fourth patient was admitted promptly but not brought to operation for 66 hours. All four patients required resection and died relatively soon after operation. A fifth patient was in the hospital, debilitated by polypoid adenomatosis of the entire gastro-intestinal tract, when

his intussusception appeared. Barium enema was not attempted. The child was too enfeebled to withstand his prompt and satisfactory resection and died two days later.

What are the reasons for the resistance among surgeons to a form of treatment which has in so many hands given results superior to those achieved by primary operation? The objections commonly raised are these: (1) The original diagnosis may be uncertain. (2) It is difficult to be sure of reduction. (3) Will not the recurrence rate be higher? (4) Polyps and other tumors cause many intussusceptions and will not be found. (5) In the unsuccessful cases a dangerous delay will have been caused. (6) The bowel may rupture. (7) Nonviable bowel may be reduced.

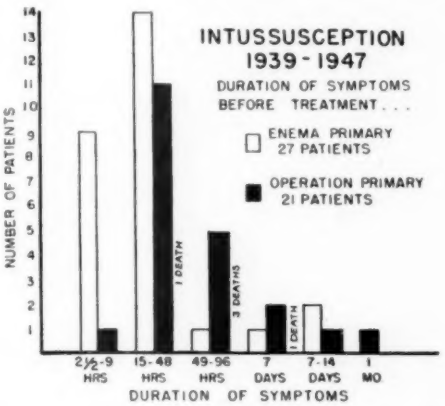

FIG. 6.—There are more patients with symptoms of short duration in the group treated by enema, but four patients in that group had symptoms of over 48 hours' duration.

To answer these objections briefly: (1) As to being sure that one is dealing with intussusception, the diagnosis by fluoroscopy is fairly simple, the criteria are characteristic, and expertness in their recognition is soon acquired. Most cases of intussusception are diagnosed clinically, but a few require barium enema for definitive diagnosis. A good many intussusceptions have been inadvertently reduced in the course of what was meant to be a purely diagnostic barium enema. (2) Accuracy of diagnosis of complete reduction is high. We have made an erroneous diagnosis of complete reduction once in 27 treatments. If there is any doubt, all that is required is a momentary inspection of the cecum and terminal ileum through a McBurney incision. Even if we were to insist on operative confirmation of every enema reduction, the procedure would still be safer and easier than operative reduction alone. With or without operation the use of hydrostatic pressure is safer and quicker than manual pulling and hauling. (3) As to the rate of recurrence, it is about 2 per cent in patients after reduction by either method. We have had one recurrence in each group in the past ten years. If there is a recurrence, the intussusception should be reduced by enema and the abdomen then explored with the expectation of finding a polyp or other causative lesion. (4) As concerns the possibility of failing to discover such a tumor by barium enema alone, the incidence of tumor is only 2.5 per cent in children under two years who suffer 70 to 90 per cent of the intussusceptions. Furthermore, the tumors which cause intussusception are rarely dangerous in themselves. Table IV shows our experience with such lesions—five polyps, two Meckel's diverticula, and one focus of ectopic pancreatic tissue. (5) As for the delay which may be caused by an unsuccessful attempt at reduction by enema, it requires less than

half an hour from start to finish to reduce an intussusception by this means. At night it is our practice to call the operating room as soon as we make the diagnosis. We usually have finished the reduction in time to cancel the operation before preparations in the operating room have been completed.

We should like to emphasize one point, that this method is not a substitute for surgery, or an escape from surgery. It is as much a surgical procedure as is reduction of a fracture by traction. The surgeon should be as ready to perform a laparotomy, if it proves necessary, as the orthopedist is to perform an open reduction. The treatment should be carried out by a surgeon and in a hospital. It is not an office procedure nor a kitchen table remedy.

As for the next objection, rupture of the bowel, this can occur with hydrostatic pressure as well as with manual reduction during operation. However, it is much less likely to occur with enema since less force is

TABLE IV.—*Demonstrated causes of intussusception in 144 cases of intussusception in infants and children treated at the Johns Hopkins Hospital from 1889 to 1947.*

DEMONSTRABLE CAUSES IN 144 CASES OF INTUSSUSCEPTION			
Cause	Patient's Age	Type (anatomic)	Acute or Chronic
Polyp	11 yrs.	Jejunal	Chronic, recurrent
Polyp	5 yrs.	2 ileal (1 compound)	Chronic, recurrent
Polyp	6½ mos.	No data	Chronic, recurrent
Polyp	1 yr., 2 mos.	Ileal	Acute
Polyp	1 yr., 6 mos.	Ileal	Acute
Meckel's diverticulum	2 yrs. 1 mo.	Ileo-colic	Acute
Meckel's diverticulum	1 yr., 2 wks.	Ileal-ileocaecal (compound)	Acute
Ectopic pancreatic tissue	10 mos.	Ileal	Acute

employed and that force is diffusely distributed. We employ three feet of pressure and never manipulate the bowel. There have been a few instances in Scandinavia of rupture from manual reduction through the intact abdominal wall with or without enema. Mortimer[19] in 1891 tested the resistance of the colon in fresh cadavers to fluid injected under pressure after the ileocecal valve had been ligated. He found serosal cracking beginning occasionally at five feet of pressure, and usually present at eight feet. The bowel sometimes ruptured at six feet of pressure, but more often at higher pressures. In this connection it may be amusing to recall the advice of Forest[20] in the New York Medical Record of 1889, that in reducing intussusception with enema one should "lay the child in the hallway and mount the stairs" holding the fluid reservoir, although he does say that after mounting to a height of 10 feet one should exercise caution![7] As for reducing nonviable bowel, it would appear that with a pressure of three feet one cannot reduce gangrenous bowel. Our experiments indicate, as others have maintained, that irreducibility of an intussusception is determined by adhesions between the sheaths and by the degree of edema. Both factors increase with time and become more effective in preventing reduction as the damage to the bowel increases.

Table V summarizes our experience in 28 dogs in which intussusception was produced by a method described elsewhere.[18] After periods of observation of 18, 28, 38, and 48 hours the dogs were again operated upon under ether anesthesia, the serosa of the undisturbed intussusception was cultured, and an attempt was made to reduce the intussusception by saline enema under direct vision with just three feet of pressure. It will be seen that reduction by enema was attempted in 28 dogs. In no case did the bowel rupture. In no case did abscess, peritonitis, or any other possible consequence of the reduction of nonviable bowel develop in any animal with a reduced intussusception. The experiments support the clinical observation that one is unlikely to reduce gangrenous bowel by a hydrostatic pressure of three feet. In several instances in another group of animals it was possible to reduce manually invaginations which could not be reduced by hydrostatic pressure.

TABLE V.—*Experimental results. Intussusceptions reduced by hydrostatic pressure at stated intervals after production. No perforation, no reduction of nonviable bowel. Pathogenic bacteria often present on serosal surfaces.*

Duration of Intussusception	Number of Dogs	Result of Enema	Fate of Dog	Peritoneal Cultures
18 hours	4	All reduced	1 anesthetic death 3 survived	3 sterile 1 positive
28 hours	3	All reduced	All survived	2 sterile 1 positive
38 hours	6	All reduced	All survived	1 sterile 5 positive
48 hours	15	2 spontaneously reduced 1 reduced with enema 12 irreducible	2 survived Survived 2 anesthetic deaths 1 survived 8 died 1 sacrificed	 2 sterile 8 positive

A few other interesting points are brought out by the study. In 7 out of 13 instances it was possible to culture pathogenic bacteria—*escherichia coli, clostridium welchii, streptococcous fecalis,* and others—from the serosal surface of viable intussuscepted bowel. This fact may well explain the high fever seen in patients after operation, and the frequency of wound infection and of postoperative adhesions.

Pathologic study of the experimental intussusceptions brought out a number of points of interest. Necrosis begins at the very tip of the intussusception and extends along the outside of the intussusceptum on the returning limb. The microscopic changes are successively those of vascular engorgement of the villi, edema of the submucosa and subserosa, hemorrhage, and finally gangrene. The inner circular muscle coat succumbs sooner than the outer longitudinal muscle coat.

In summary: The cumulative experience of 75 years in the treatment of intussusception shows reduction by hydrostatic pressure to be a safer primary

approach than operation. This conclusion is supported by our clinical and experimental observations. In 27 patients treated primarily by barium enema there were no deaths. The morbidity and length of hospital stay were much reduced when compared with those of patients treated primarily by operation during the same period. The use of hydrostatic pressure under fluoroscopic control in the treatment of intussusception is auxiliary to surgery and not in opposition to it.

Author's Note: Since this paper was submitted for publication 6 additional children with intussusception have been treated by barium enema reduction, with no deaths. In 3 enema alone sufficed, in 3 a McBurney incision was required to complete reduction at the ileocecal valve. There is thus a total of 33 cases of intussusception treated primarily by barium enema, with no deaths, 24 of the number being reduced by the barium enema alone.

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PRIMARY "INFLAMMATORY" CARCINOMA OF THE BREAST*

A Report of Five Cases and a Review of the Literature

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THE BREAST is the site of a number of varied pathologic lesions, but none more interesting than "Inflammatory Carcinoma of the Breast," so named because clinically the breast presents signs commonly associated with inflammation.

Frequently these skin manifestations are misdiagnosed and treated as a dermatological or inflammatory lesion rather than as a neoplasm. Four patients with inflammatory carcinoma treated on the general surgical service of the State University of Iowa Hospitals in the last year had been treated as having acute inflammatory lesions prior to admission. To create awareness of this unusual type of neoplasm, and thus prevent loss of valuable time in the institution of therapy at the onset of the disease, available information pertaining to "inflammatory carcinoma" is presented. The material has been obtained from a review of the literature and from personal observation of five patients treated for inflammatory carcinoma at this hospital during the past year.

Two clinical varieties of inflammatory carcinoma of the breast exist. In one group designated as primary, the inflammatory signs arise simultaneously with the carcinoma in the skin of a previously normal breast. It is to be emphasized that in this group there is usually no mass present in the breast, the absence of which frequently accounts for the delay in the correct diagnosis. In the group designated as secondary, the inflammatory manifestations may appear suddenly in a breast which has long been the seat of a scirrhous carcinoma, or in the mate of such a breast or it may follow mastectomy for carcinoma either at the original site, or in the opposite breast. The clinical course and behavior of both types is very much the same. This paper is confined to a discussion of primary inflammatory carcinoma.

Inflammatory carcinoma of the breast presents definite clinical and pathologic characteristics. It is an unusual type of malignancy in that in the early stages it presents signs and symptoms usually associated with acute or subacute inflammation. It is a type of neoplasm which presents a rapid rate of spread with early metastasis, and a discouraging response to all forms of treatment.

HISTORICAL

Probably the first description of this type of mammary carcinoma was by Volkmann[20] in 1875, who called it "Mastitis Carcinomatosa." Since then this neoplasm has received a variety of names, such as Carcinoma erysipeloides,[4, 16] erysipelas carcinomatosa,[10] telangiectatis carcinoma,[21] malignant lymphangitis,[5] and acute carcinoma,[9] usually depending upon the author's interpretation of the signs, symptoms and pathological characteristics. The designation of

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"Inflammatory Carcinoma" was first suggested by Lee and Tannenbaum[11] in 1924 in a report of 28 cases. This terminology has also been used by Taylor and Meltzer[19] who in 1938 presented 38 cases, and by Fox.[8, 9]

Because of the clinical characteristics presented by this neoplasm the nomenclature most favored is "Inflammatory Carcinoma."

INCIDENCE

Fortunately this neoplasm is infrequent in occurrence, representing but 1.3 per cent of all cases of mammary cancer, according to Lee and Tannenbaum,[17] Taylor and Meltzer,[19] give a somewhat higher figure of 4 per cent. At the University of Iowa Hospitals five patients with inflammatory carcinoma have been treated in the past 12 months and have constituted 4.2 per cent of malignant breast lesions seen during that period.

The age incidence of inflammatory carcinoma parallels that of breast carcinoma in general. In the 28 cases reported by Lee and Tannenbaum[17] the average age was somewhat younger, being 44.5 years. Ten of the cases, or 39 per cent, occurred in women under 40 years of age. The youngest patient reported with inflammatory carcinoma was 24 years old. Five cases treated at this hospital presented an average of 56 years.

PATHOLOGY

Early workers, impressed by the gross inflammatory signs, tried to read inflammatory features into the microscopic picture of the disease. Schumann[18] in 1911 attributed a "toxin-forming power to the carcinoma cells which by setting up a violent irritation, produces a round-cell infiltration closely simulating a primary mastitis."

Learmonth[13] also was of this opinion when he said: "We have to deal with a highly virulent type of cancer cell which is capable of engendering a marked inflammatory reaction."

As early as 1889 Bryant[3] understood that the inflammatory signs of this clinical form of breast carcinoma were due to extensive lymphatic invasion by cancer cells producing edema and capillary congestion. Some early investigators held that the tumor spread was chiefly through the subepidermal capillaries and venules. Ewing[6] advanced the opinion, currently shared by Case[5] and others, that the disease was primarily a continuous growth of carcinoma through the lymphatic vessels.

Because of uncertainty as to whether the vessels by which the tumor cells spread were lymphatics or capillaries, Camiel and Bolder[4] studied approximately 200 serial sections of biopsy material. Barring artefaction the authors presumed the presence of an erythrocyte in a vessel might be taken as evidence of a capillary or venule. None were found.

Using this same criterion, skin biopsies taken from breasts of four of the patients with inflammatory carcinomas treated at the State University of Iowa Hospitals showed many red blood cells in the vessels which contained tumor cells. This is illustrated in the photomicrograph shown in Figure 1. It seems reasonable that spread of the tumor cells may be either by the lymphatics or

capillaries; the disease being primarily a continuous growth of carcinoma through the lymphatic vessels with concomitant blood vessel invasion and congestion of the subpapillary capillaries and venous plexus.

It should be emphasized that true inflammation does not exist. There is no body reaction to an irritant, but the extensive lymphatic blockage causes edema,

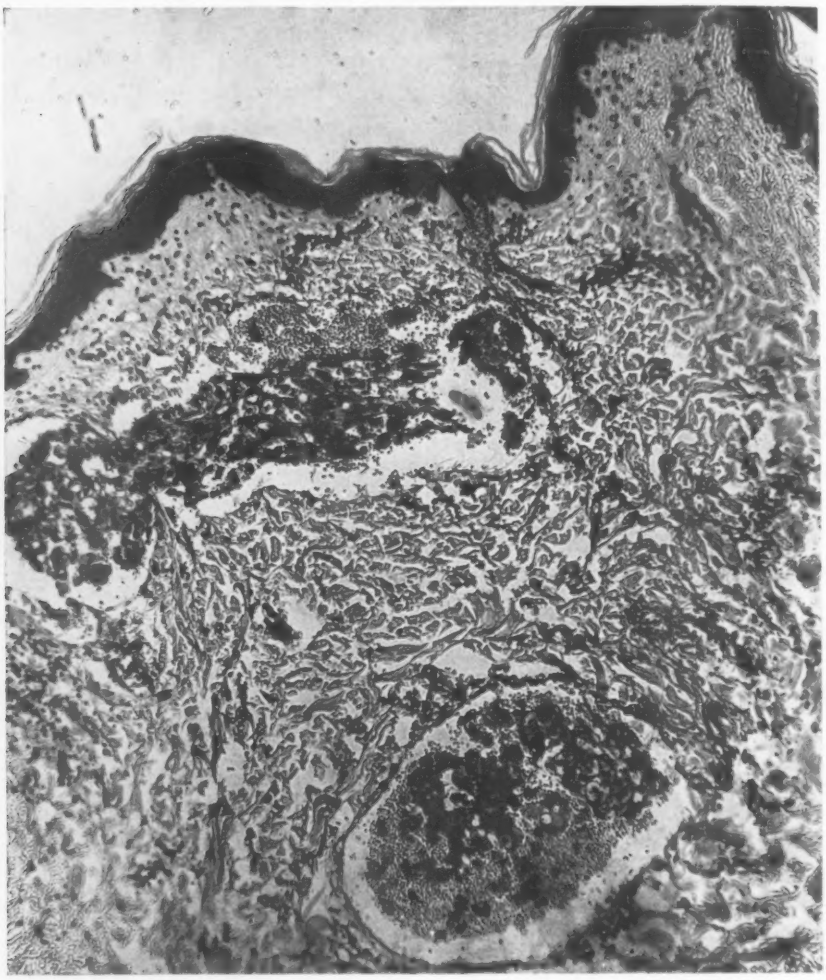


FIG. 1.—Photomicrograph showing evidence of spread of inflammatory carcinoma by blood vessels. Note erythrocytes in vascular space with neoplastic cells.

and the capillary congestion produces marked reddening and heat. The round cell infiltration often noted is also due to blockage of lymphatics,[12] and not to an inflammatory reaction.

The histologic type is usually an undifferentiated carcinoma. Microscopic study shows nests of neoplastic epithelial cells in the corium. The neoplastic cells are usually collected in the vascular spaces, lymphatics or both. The spread of the carcinoma is subdermal rather than intradermal, an important

differential point from Paget's disease which involves primarily the epidermis. (Fig. 1.)

CLINICAL SYMPTOMS

In the majority of instances the initial symptoms are subjective in nature. The patient will complain of heaviness or aching of the breast. The pain will vary from a dull ache to an intermittent shooting pain.

The patient next notices skin discoloration which varies from bright red to purple and which varies in size and shape. At this time an increase in the size of the breast is also noted. If the patient consults a physician at this stage of the disease the clinical picture closely resembles an acute inflammatory process and is usually treated as such. Of the five patients treated at the University of Iowa Hospitals in the last year, four had received treatment for acute mastitis before being referred for treatment. In the cases reported by Lee, over one-half had been operated upon with a diagnosis of chronic or subacute abscess.

A great amount of valuable time is lost when the neoplasm is treated as an acute inflammatory process. Reuter and Nomland[17] recommend that inflammatory signs developing in the skin or about the breast which do not subside within two or three weeks warrant an incisional biopsy for diagnosis. This is sound advice and if followed it will prevent unnecessary delay in the patient receiving prompt treatment.

Coincident with color change there is an increase in the size of the breast. The involved breast is usually two or three times the size of the normal breast. Patients with inflammatory carcinoma also complain of weakness, easy fatigability, and weight loss, symptoms which may be associated with any malignancy or chronic illness.

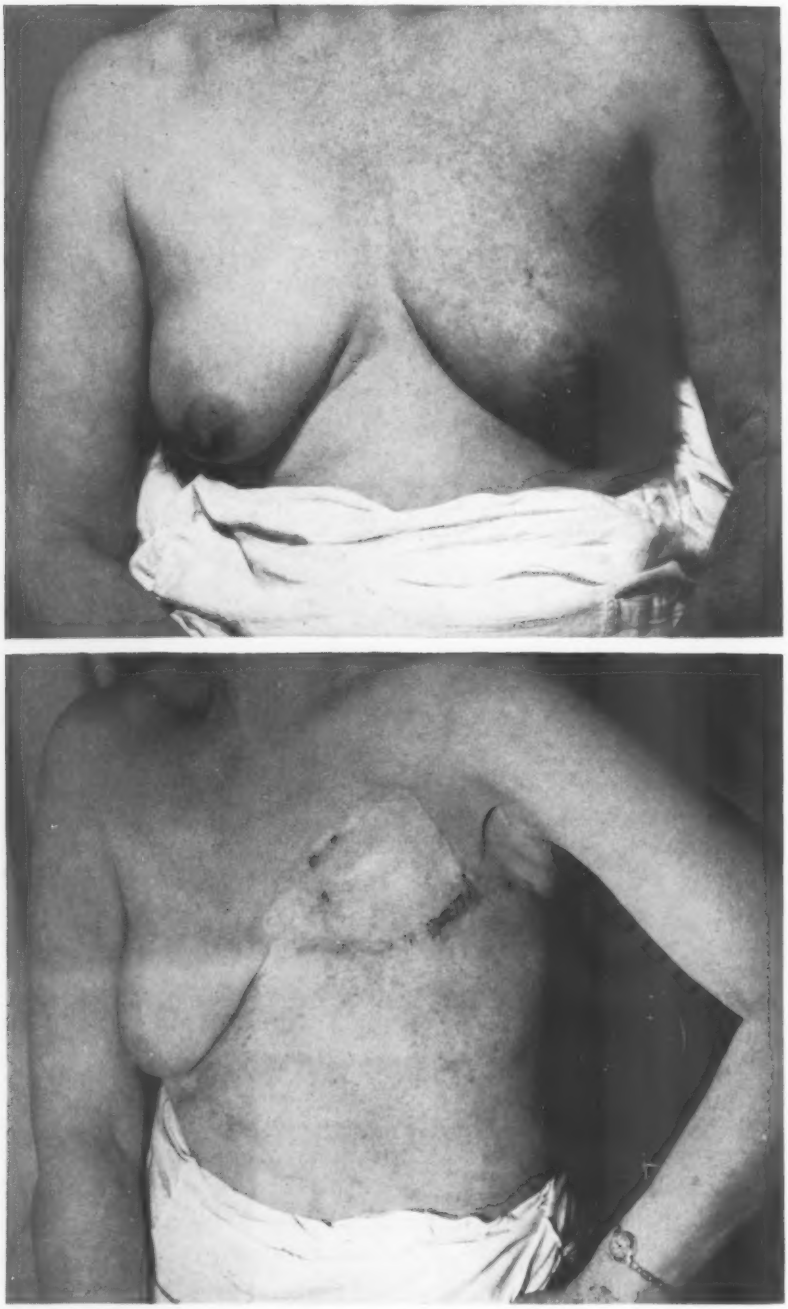
As a general rule the patients do not present an elevation of temperature. There are no constant findings in the microscopic examination of the blood. Leucocytosis is the exception rather than the rule. A secondary anemia may be present.

CLINICAL SIGNS

Examination of the involved breast in the early stage shows areas of skin discoloration. The color is usually red or reddish purple, but occasionally may be reddish brown in appearance. This discoloration may be seen as irregular patches, or as streaking, the latter resembling lymphangitis in appearance. As the disease progresses the skin becomes deep red or reddish purple in color and begins to extend beyond the confines of the breast. The skin becomes pitted and edematous, and there is pronounced induration of the skin. At this stage the skin shows the "orange peel" or "pitted" appearance. In some cases nipple retraction is present.

The involved breast may be enlarged two or three times the size of the other breast. At the outermost area where involved skin and normal skin merge there is an irregular raised periphery and the appearance is very similar to an acute inflammatory lesion such as erysipelas.

A



B

FIG. 2(A)—Left breast showing irregular erythematous skin discoloration in early stage of primary inflammatory carcinoma. Note resemblance to acute lymphangitis. (B)—Patient shown in 2A after intensive preoperative Roentgen therapy and radical mastectomy. Note diffuse spread of lesion and the erysipelas-like border at the periphery.

The skin is warm to touch and not infrequently tender. Usually a palpable mass is not present.

The gross appearance of inflammatory carcinoma of the breast and the rapid progression of this disease is well illustrated by Figures 2, 3, 4, 5 and 6.

DIAGNOSIS

In establishing a diagnosis the following diseases must be differentiated from primary inflammatory carcinoma:

True inflammation. As previously mentioned a major difficulty in the diagnosis of inflammatory breast cancer exists in the fact that in its early stages the disease may closely resemble a true inflammatory process of the mammary gland, such as an abscess or an acute mastitis. The skin overlying an abscess of the breast is apt to be smooth, shiny, and tender. Leucocytosis, fever, and other general signs are usually present. Inflammatory carcinoma is typically nontender, covered with edematous "orange peel" skin and unassociated with systemic fever or leukocytosis. An induration in the breast due to lactation mastitis should show in a few days either evidence of resolution or the clinical appearance of abscess. Any case of supposed mastitis persisting longer than two weeks calls for the provisional diagnosis of cancer and warrants incisional biopsy.

Erysipelas. The differential diagnosis between erysipelas and inflammatory carcinoma may be impossible on the basis of gross appearance. However, in this region erysipelas is rather uncommon. The disease is quite acute and prostration is usually striking. High fever and marked leucocytosis are present. The lesion is vivid red, disappears on pressure and is not particularly sensitive. The acute history, with the constitutional reaction and dramatic spread of the lesion, together with the decided increase in leucocyte count is sufficient to distinguish erysipelas from inflammatory carcinoma.

Paget's disease of the nipple. The skin lesion in Paget's disease involves the nipple, areola, and surrounding skin and presents a reddened, granular surface discharging a yellow viscid fluid. At times the appearance is that of chronic eczema with encrustations. The lesion is frequently confined to the areola alone and the nipple disappears as the disease progresses. In inflammatory carcinoma the nipple is usually not involved other than showing evidence of retraction. There is more diffuse discoloration extending out from the nipple. Biopsy of the involved skin is of value in the final differentiation. The spread of Paget's disease is intradermal, while inflammatory carcinoma is subdermal. Typical signet ring cells known as "Paget Cells" are also seen.

Plasma cell mastitis. In the acute phase of plasma cell mastitis the signs and symptoms of a diffuse inflammation radiating from the nipple are seen. However, it varies from inflammatory carcinoma in that the inflammation will subside in from a few days to a week. After subsidence of the acute manifestations a firm irregular tumor remains. With inflammatory carcinoma, as has been mentioned previously, there is usually no mass palpable. Histologic examination of the skin in plasma cell mastitis shows absence of carcinoma,

A

B

FIG. 3(A)—Breast showing skin discoloration and enlargement of primary inflammatory carcinoma. Note small area of involvement on opposite breast. (B)—Patient shown in 3B after intensive Roentgen therapy which had no effect in preventing progression of lesion.

and examination of the mass itself shows the typical picture of plasma cell mastitis as described by Parsons, Henthorne, and Clark.[14]

Tuberculosis of the breast. In this condition the onset is less acute. A tumor is present in the breast which is soft in consistency. Early in the course, redness of the overlying skin appears and later areas of softening. As the process extends the skin over the softer portions thins out and finally sinuses form which are usually multiple. Physical signs of tuberculosis elsewhere may also be demonstrated. This condition, from its chronicity and physical signs, may be readily distinguished from inflammatory carcinoma.

Lymphoma. Hodgkins disease can produce a picture indistinguishable from inflammatory carcinoma of the breast. The mechanism is undoubtedly a similar one, with acute diffuse lymphatic blockage by multiplying malignant cells. Biopsy is necessary to establish the diagnosis.

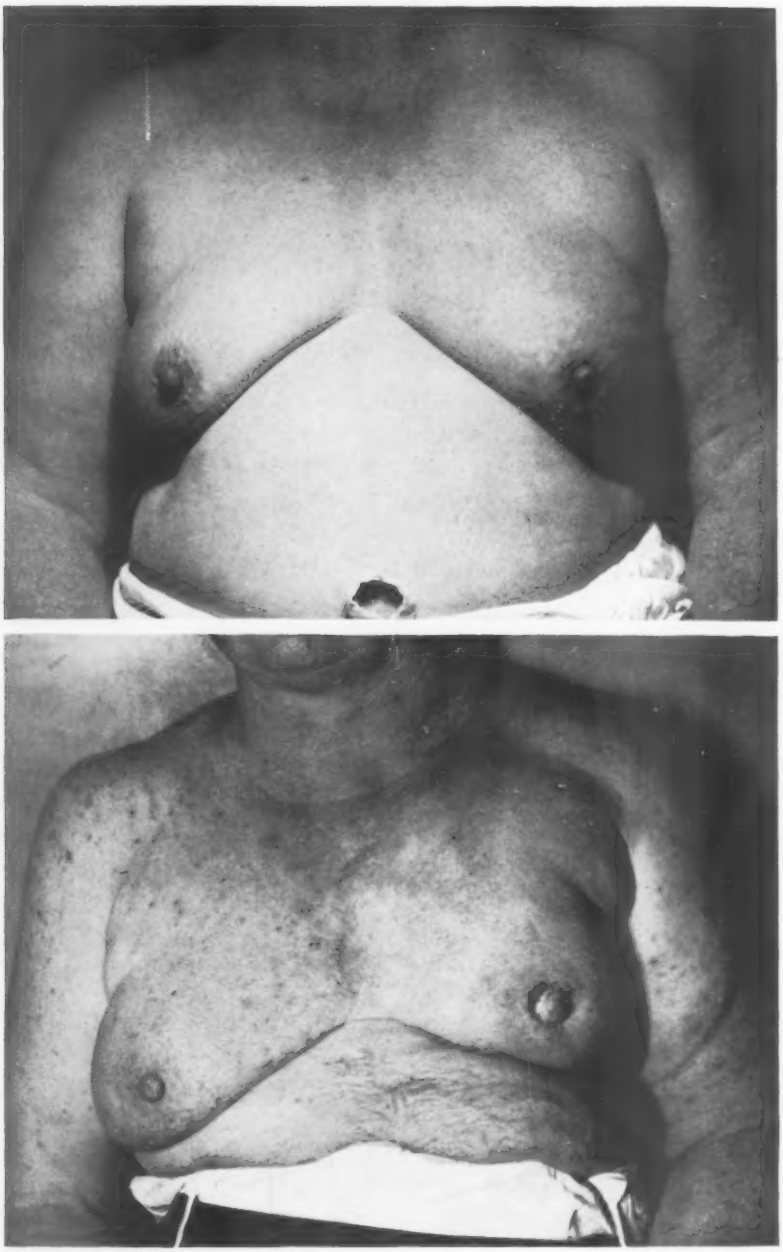
Metastasis. It has been previously mentioned that the local lesions of inflammatory carcinoma spread with striking rapidity. Metastasis also occurs early in primary inflammatory carcinoma of the breast, involving, in practically all cases, the axillary or supraclavicular nodes. Taylor and Meltzer presented 25 cases of primary carcinoma. In 11 of the 25 cases medical advice was sought within three months of the onset of symptoms, seven saw a physician within six months, and all but one within a year. All 25 cases showed axillary involvement, and nine cases in addition showed clinical evidence of visceral metastases (liver or lung) or bone metastases at the time of examination. At the University of Iowa Hospitals the five cases described below presented themselves for treatment at three months, five months, nine months, 18 months, and 12 months respectively. Four of the five patients presented axillary metastasis. Two in addition evidenced supraclavicular matastases. Liver metastasis was present in one patient. The opposite breast is also frequently involved.

Treatment. Whatever the form of therapy the outlook for the patient with inflammatory carcinoma is discouraging. As far back as 1814 Bell[1] appreciated the gravity of these inflammatory signs when in a discussion of carcinoma of the breast he said "When a purple color is on the skin of the tumor, it is a very impropitious beginning."

Portmann,[15] after an extensive review of the treatment of breast carcinoma, stated that brawny red induration and inflammation of the skin, and edema as evidenced by orange peel skin were to be considered as criteria of incurability. Both of these are present in inflammatory carcinoma. Metastasis takes place very early in the disease. In addition the histologic type reveals the carcinoma to be highly malignant. For these reasons the response in the majority of cases to the usual forms of treatment, namely Roentgen-rays, surgery or a combination of the two, has been uniformly bad.

Taylor and Meltzer report 19 cases treated by Roentgen-rays. Three patients were still alive at the end of six, ten and twenty-five months. Nineteen patients lived two to nineteen months after institution of treatment with an average duration of life of 9.2 months.

A



B

FIG. 4(A)—Left breast showing the early skin changes and enlargement of primary inflammatory carcinoma. (B)—Patient shown in 4A after intensive Roentgen and testosterone therapy. Acne and hirsutism due to the testosterone.

Learmonth,[13] reports one patient, treated by surgery, alive after seven years. Brackertz,[2] recommended surgical excision, allowing a 10cm. margin to every trace of redness. Lee and Tannebaum report six patients treated by surgery who had an average duration of life of one year and nine months. Their patients all did poorly, developing prompt evidence of axillary or supraclavicular disease, skin recurrence or involvement of the opposite breast. Fox,[8, 9] reported uniformly bad results with operative treatment.

The present series of five cases illustrates the difficulties presented in arriving at a definite conclusion as to the most suitable type of treatment.

Case 1.—A 61-year-old white female complained of aching of the left breast with accompanying discoloration of the skin of three months duration. During this time the patient had received treatment for acute mastitis.

Physical examination revealed the right breast to be two times the size of the left breast. The skin of the breast showed an irregular reddish-purple discoloration. Metastatic nodes were present in the right axilla and supraclavicular regions. Biopsy of the skin was reported as undifferentiated carcinoma. Because of metastases, roentgen therapy was chosen as the method of treatment. This, however, was of no apparent benefit, and the lesion showed a steady progress in severity involving the skin of the back, abdomen, and opposite breast. This patient at last report was rapidly losing ground, 13 months after the onset of her disease.

Case 2.—A 58-year old white female complained of a "red, itchy, sore left breast" of nine months duration. During this time the patient had been treated for acute mastitis.

Physical examination revealed the left breast to be enlarged one and one-half times that of the right breast. The skin of the breast was indurated and several irregular erythematous areas were present. Axillary metastasis was present.

The patient received an intensive preoperative course of roentgen therapy. Examination two months later showed definite regression of the lesion, so much so that a radical mastectomy was performed. Histologic examination of the lesion was reported as poorly differentiated carcinoma. This patient did extremely well for approximately nine months after discharge from the hospital. At this time she developed a purpuric lesion of the anterior and lateral chest wall. She was treated by her local physician for herpes zoster. When the patient returned to this hospital for examination, biopsy revealed the lesion to be carcinoma. Testosterone as a palliative resort was started within the last week; evaluation of the treatment cannot therefore be made at this time.

This patient of the five patients presented had shown the best response to treatment having been free of evidence of carcinoma approximately one year. However, with evidence of recurrence of the carcinoma now present it illustrates the discouraging response of inflammatory carcinoma to available therapy.

Case 3.—A 58-year-old white female complaining of enlargement and irregular erythematous discoloration of the left breast of one years' duration. Physical examination revealed the left breast to be two to three times the size of the right breast. There was a diffuse erythematous discoloration and lymph-edema of the breast. Nipple retraction was present. A metastatic node was palpable in the left axilla.

The same type of therapy was decided upon as was given in Case 2. The patient received an intensive course of roentgen therapy. The patient returned in two months for radical mastectomy. Examination revealed an irregularity of the right tenth rib anteriorly. To rule out a metastatic lesion this area was explored. This revealed a fracture of the tenth rib at the costo-chondral junction which accounted for the irregularity palpated. Through this incision the liver was felt through the peritoneum and found to be nodular. A small incision was made through the peritoneum and many metastatic nodules were found. Biopsy revealed the presence of carcinoma. Testosterone

FIG. 5

FIG. 6

FIG. 5.—Left breast showing characteristic enlargement that occurs in primary inflammatory carcinoma.

FIG. 6.—Right breast and surrounding skin showing marked involvement with primary inflammatory carcinoma. This patient had received intensive Roentgen therapy.

therapy was instituted without benefit. The patient died several months after discharge from the hospital approximately 19 months after onset of symptoms.

Case 5.—A 55-year-old white female complaining of soreness and skin discoloration of the left breast of five months duration.

Physical examination revealed the left breast to be enlarged two times that of the right breast. The skin of the breast was diffusely reddened and indurated. Axillary and supraclavicular node metastases were present. Biopsy of the skin and of a supraclavicular node were reported as undifferentiated carcinoma.

The patient was given an intensive course of roentgen therapy. Examination two months later revealed progression of the neoplasm. Testosterone therapy was started. A dosage of 100 mg. daily was prescribed. The patient has returned for examination at intervals of one month for the past eight months. At the completion of two months of therapy there was evidence of regression of the inflammatory signs with an increased feeling of well being experienced by the patient. This improvement continued for an additional three months. At this time examination once again revealed definite evidence of spread of the carcinoma. At the last examination eight months after testosterone therapy was instituted the patient has continued to experience a feeling of well being in spite of progress of the local lesion. It is believed that this patient has received definite benefit from testosterone therapy.

Case 5.—A 52-year-old white female admitted with diabetic gangrene of the right foot of four weeks duration. In addition physical examination revealed a markedly enlarged right breast. The skin was brawny reddish purple in color, and indurated, presenting an orange peel appearance. The skin involvement extended to the supraclavicular lesion superiorly, the medial portion of the left breast, the costal margin inferiorily, and the posterior axillary line posteriorly. Skin biopsy revealed poorly differentiated adenocarcinoma.

The patient gave a history of the breast lesion beginning approximately 18 months earlier. It was ignored until two months prior to admission at which time a physician was consulted and an intensive course of roentgen therapy was given. From the appearance of the lesion it appeared to have been benefitted very little by this treatment.

The patient was seen by the Department of Rontgenology who expressed the opinion that further therapy would be of no value. Testosterone therapy was prescribed. The patient died suddenly 11 days after therapy was started and one week after discharge from the hospital. This patient survived 19 months after onset of her symptoms.

The above case histories do not lead to any definite conclusions about the treatment of inflammatory carcinoma other than that the outlook is very discouraging. Three patients have died 13, 19, and 19 months respectively after onset of their disease. Two patients are still alive 19 and 21 months after onset of symptoms. However, continued spread of the carcinoma in these cases, in spite of all available therapy, suggests an apparently hopeless prognosis.

The most favorable result was obtained in Case 2, the patient receiving preoperative roentgen-ray followed by radical mastectomy, although recent examination showed evidence of recurrence. Roentgen-rays alone have proved to be of little benefit in stopping the spread of inflammatory carcinoma, other than showing an initial favorable response in Case 2.

Case 4 is of interest in the systemic improvement experienced by the patient when placed on testosterone therapy. In spite of continued progress of the local lesion the patient continued to experience a feeling of well being and was able to carry on her normal household duties.

In view of the highly malignant nature of this disease, it is recommended that the patient be given every opportunity for cure through a combination of

all therapy available. This would include an intensive course of preoperative roentgen-ray therapy, followed by radical mastectomy in cases deemed operable. The inoperable cases who do not respond favorably to roentgen-ray therapy should be given the benefit of testosterone proprionate. The greatest aid for improvement of therapy would be earlier diagnosis. Recognition of the disease at the onset would allow prompt institution of treatment and perhaps produce a more favorable result.

SUMMARY

1. Primary inflammatory carcinoma, an interesting, infrequent and highly malignant tumor of the breast has been described.

2. The age incidence parallels that of breast carcinoma in general.

3. The disease is primarily a continuous growth of carcinoma through the lymphatic vessels with concomitant blood vessel invasion.

4. The histologic type is usually undifferentiated carcinoma or poorly differentiated adenocarcinoma.

5. Symptoms are acute in onset, most frequently characterized by enlargement and aching of the breast with concomitant inflammatory signs, and, in the early stages of the disease resembling an infectious process.

6. Differential diagnosis is made by absence of generalized symptoms indicating an infectious process, normal blood count and biopsy.

7. Metastasis takes place early and in practically all cases the axillary and supraclavicular nodes are involved.

8. Response to treatment is uniformly poor. Roentgen therapy and surgery in the operable cases has been of some value. Roentgen therapy and testosterone proprionate are recommended for the inoperable cases.

9. Biopsy of inflammatory lesions of the breast which persist over two weeks, to aid in early diagnosis and institution of proper therapy, is urged.

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THE SILENT GALLSTONE: A TEN TO TWENTY YEAR FOLLOW-UP STUDY OF 112 CASES*

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BECAUSE, SINCE THE ADVENT OF CHOLECYSTOGRAPHY, the problem of the silent gallstone has become an increasingly frequent and, consequently, an increasingly important one, several pertinent questions are being asked. How often are gallstones silent and how often is the physician confronted with the problem of the silent gallstone? What advice should be given the patient who has silent gallstones? Should he be told to undergo cholecystectomy or should he be told to await development of dyspepsia or colic before submitting to surgical treatment? Should he be urged to undergo cholecystectomy after the first attack of colic or after several attacks have occurred?

FREQUENCY OF THE SILENT GALLSTONE

The frequency with which the silent gallstone occurs in the general population is not known. Surveys using cholecystography, so far as we can find, have not been made to determine the incidence. Some idea of its frequency, however, may be gained by consideration of data obtained from necropsy studies. Robertson,[1] in reviewing the records of 1,027 cases in which necropsy was performed and in which gallstones were or had been present, found that in 61 per cent there had been no suspicion by the patient or his attending physician that gallstones did exist. Obviously, given the opportunity of careful questioning by the physician during the patient's life, not all of the 61 per cent would have been classified as cases of silent gallstones but, after taking into consideration this source of error, it seems that the percentage of silent gallstones may well be high. When these data are considered in relation to the finding of Robertson and Dochat[2] that in 16.3 per cent of 16,926 necropsies gallstones were present and that the proportion ranged from 0.1 per cent in the first decade of life to 32.7 per cent in the ninth decade of life, one is impressed not only with the large number of gallstones but also with the large number of silent gallstones potentially present in the general population.

PREVIOUS VIEWS ON TREATMENT FOR THE SILENT GALLSTONE

There is widespread agreement that surgical treatment is indicated when calculous disease of the gallbladder produces distressing dyspepsia or colic

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and when the complications thereof arise, but agreement that silent gallstones require surgical treatment is lacking. This has been true both in the early years and since cholecystography has permitted such a diagnosis frequently. Before 1924, the remarks of Kehr,[3] W. J. Mayo[4] and Osler[5] summarize the several attitudes of the day toward the silent gallstone. In 1901 Kehr stated that "quiet lying gallstones are no subject for treatment, for a persistent latent stage is almost as good as a cure." On the other hand, in 1911, W. J. Mayo said: "Ten years ago we heard a great deal about 'innocent' gallstones which meant that gallstones existed without symptoms and that their presence was not suspected until postmortem examination brought them to light. . . . That temporary palliation can be procured with nonoperative treatment cannot be denied, but the cure of the patient can only be brought about by surgical means." Osler[5] in the following year, 1912, said that surgical treatment of the gallbladder was indicated "after repeated attacks of colic, in the presence of a distended gallbladder associated with attacks of pain or fever, and a stone in the common duct."

Since 1924 and the introduction of cholecystography by Graham and Cole the same variations in attitudes toward treatment of the silent gallstone may be found. White,[6] in 1928, stated that "many silent gallstones show a gallbladder wall that is bacteriologically negative. Cholesterol stones especially lie dormant for years. Surgery should be based on symptoms, not on the mere presence of stones or low grade infection. I believe that most patients escape perceptible injury to other organs." Mason and Blackford[7] said in 1932: "We fear that occasionally surgical relief is advised because of refinements in the interpretation of symptoms together with new methods of diagnosis of gallbladder disease, rather than because of the former clinical symptoms." Musser[8] concluded in 1934 that "it is doubtful if interference is indicated for the first or even the second biliary colic if evidence of infection is lacking." Andrews[9] said in 1936, "I think the operation on silent gallstones is a scandal."

On the other hand, Lahey[10] in 1938 expressed the conviction that there are no harmless gallstones "and that because of the above dangers [acute cholecystitis and biliary obstruction] all gallstones should be removed. It has frequently been urged and practiced that a proper attitude to take toward gallstones is that operation should not be undertaken until they produce symptoms. If one deals with gallstone patients in large numbers one cannot help being impressed with the fact that this philosophy results in many situations and in many fatalities which would not have occurred had the operative procedure been undertaken earlier." Cheever[11] shares Lahey's opinion that "there are no harmless gallstones . . . that cholecystectomy should be advised unless special contraindications exist when the diagnosis of gallstones is made." Carter, Greene and Twiss[12] in 1939 said that "in every instance in which stones can be demonstrated to be present, surgery is indicated to prevent the dangers of acute ulcerative cholecystitis."

Fitz[13] in 1942 expressed the belief that the "most judicious therapy of 'silent' gallstones may be different than in their later stages of symptomatic activity. . . . The very ease with which gallstones can now be recognized

by cholecystography may lead to errors in therapeutic judgment. . . . It is not harmful to follow patients with 'silent gallstones' for a time before advising surgery, and it may be helpful."

It is interesting to compare the views on this problem as stated in "Nelson's new loose-leaf surgery" and "Nelson's new loose-leaf medicine." Whipple,[14] writing in the former, concluded that "if the gallbladder is the seat of definite chronic inflammation and especially if the chronic inflammation is associated with gallstone formation, surgery is the only therapy that offers permanent and satisfactory cure." Writing in the latter, Miller and Machella[15] stated that "opinion is divided as to the propriety of operating in cases of stones in the gallbladder without symptoms and in which the diagnosis is made accidentally. Since such patients have gone through a long life without digestive symptoms, one would seem to be justified at least in postponing surgery until some symptoms develop."

Meakins'[16] stand on this question should be considered. Concerning gallstones, he expressed the belief that "their removal by operation is not indicated for the first, second, or even several attacks of colic. There may be lengthy intervals and the disability of an occasional attack may be relatively insignificant compared to the risk of operation, which is not inconsiderable, particularly in elderly people affected with diabetes, obesity, hypertension, and myocardial degeneration, all or any of which are commonly associated with gallstones." Christian,[17] in Osler's "The Principles and Practice of Medicine," expressed a view similar to that of Meakins: "The decision for or against surgical treatment should be made in the course of repeated attacks; the single attack requires operation only in the unusual case of steadily increasing severity and evidence of severe infection. There are many patients in whom the symptoms are slight, for these surgical treatment is not indicated. . . . It is rare that cholecystectomy is advisable for the first attack, even though it is severe and typical. The present author has long followed the policy just stated; in his patients it has led to no serious consequences that would have been avoided by early cholecystectomy. . . ."

Watson,[18] writing in Cecil's "A Textbook of Medicine," introduced another factor. He wrote, "In the usual case, however, cholecystectomy is indicated because of the many serious complications which gallstones may produce. . . . Furthermore, the important relation of cholelithiasis to the occurrence of primary cancer of the biliary tree must be taken into account."

Finally, the opinion of Clute and Kenney[19] (1944) is that "in the great majority of simple or silent gallstones an early elective cholecystectomy is wiser than an operation into which one is forced by the complications of the disease."

The wide divergence in opinion regarding the treatment of the silent gallstone probably arises from the lack of accurate data pertaining to the incidence of dyspepsia and colic and of the complication of calculous disease of the biliary tract in cases of gallstones which are silent when discovered. Data regarding the incidence of the several complications of calculous disease of the gallbladder in large series of patients coming to operation are available

but these data do not apply to the incidence of the several complications in cases of silent gallstones.

It is doubtful whether carcinoma of the gallbladder should be emphasized as a hazard of silent gallstones adequate to demand surgical intervention for its prevention. Not only is carcinoma of the gallbladder relatively rare but also it appears that this neoplasm is found most frequently when symptoms of cholecystitis have been present for many years. Jaguttis,[20] in 1926, reported that cancer of the gallbladder developed in five of his 114 patients who had had symptoms of cholecystic disease for 10 to 25 years. Vadheim, Gray and Dockerty[21] found that symptoms of disease of the biliary tract had been present in their 77 cases of carcinoma of the gallbladder for an average of 14.2 years in 80 per cent of the patients.

Similarly, the incidence of stone in the common duct appears to be related to the duration of symptoms. In a study by Heyd[22] of 1,270 cases in which symptoms of gallstones had been present for two years or less, the incidence of stones in the common duct was 1.9 per cent. In another group of patients in which the symptoms of cholecystic disease had been present for 10 to 25 years, Heyd found the incidence of stones in the common duct to be 16.0 per cent. So long as the stone remains silent, the chances are that its site in the common duct must be small.

DATA OF VALUE IN DETERMINING POLICY ON TREATMENT

To decide for or against surgical treatment of silent gallstone, the physician should have certain information. He should know in how many cases of silent gallstone dyspepsia, and in how many cases colic, appears at a later date. He also should know in how many the complications of calculous disease of the gallbladder develop. Also, it would be of value to know how frequently complications occur with the first attack of colic. Such data would be valuable in establishing the risk to the patient who has silent gallstones of deferring surgical intervention until the first attack of colic, because the mortality of cholecystectomy for uncomplicated disease of the gallbladder is about 0.5 per cent and for complicated disease of the gallbladder, 3 per cent in the hands of an experienced surgeon. These data are needed for evaluation before one is able to decide whether or not a patient who may never have incapacitating dyspepsia, an attack of colic or complications, should undergo a surgical procedure which carries a risk of at least 0.5 per cent.

MATERIAL AND METHODS OF STUDY

In the hope of supplying some of the data not now available but necessary for decision regarding treatment of the silent gallstone, a long-term follow-up study of all cases in which gallstones were found incidentally during the course of some other abdominal operation at the Mayo Clinic was carried out. The records of 998 such cases occurring from 1925 through 1934 were reviewed. Approximately a half of these were discarded as unsuitable for long-term follow-up because the operation had been for cancer. Many others, including those with duodenal ulcer, were discarded because some of the abdominal

symptoms might have been due to the cholelithiasis. Follow-up letters were sent to 184 persons. Replies were received from 115 (62 per cent). Each patient was asked if indigestion or colic had preceded the discovery of the gallstones and several replied that symptoms had been present, so that these, too, were discarded. Finally, 112 cases were considered suitable for study. The average age of the 112 patients was 48.2 years when the gallstones were discovered.

RESULTS OF STUDY

The first report on the course of the calculous disease of the gallbladder in these 112 patients will be given.

In 30 cases indigestion developed and it is assumed that the indigestion in each case was due to the disease of the gallbladder. Under the term "indigestion" are included those cases in which such symptoms as gaseous indigestion, intolerance to certain foods, and heartburn developed, as the only abdominal distress. The severity of the indigestion varied; some patients were bothered occasionally, a few were able to control all symptoms by diet while others felt that the indigestion was incapacitating.

Twenty-one patients reported colic; more than half had experienced more than one attack of colic, but several who had had frequent and severe attacks of colic found that sooner or later the attacks had subsided so that they had had no symptoms whatsoever for years. Five of the 21 patients had both colic and jaundice; of the five, four had had slight jaundice which followed an attack of colic and was transient only. The fifth patient underwent operation immediately after the first attack of colic and jaundice.

In a total of 51 (45.5 per cent) of the 112 cases symptoms developed. In 24 of these 51 cases cholecystectomy was performed. Three of the 24 patients died postoperatively. The death of the patient who underwent operation at this clinic was due to pulmonary embolism. Of the two who underwent operation elsewhere, one death was reported by the patient's relatives to be due to subphrenic abscess and the other to a "weeping" liver. In 61 (54.5 per cent) of the 112 cases abdominal symptoms did not develop before death or have not developed since the discovery of the gallstones.

It is of interest that 28 of the patients have died. Of the 28, 21 did not, so far as can be ascertained, experience symptoms prior to death but seven did so. The cause of death was unknown in four cases, while death was due to cancer in six, to heart disease in five, to infectious disease in five, to cerebral accident in four, to amputation of the leg in one, and followed cholecystectomy in three. In not one of the six cases in which the patient died of cancer was the gallbladder the seat of the cancer.

COMMENT

Although we recognize the limitations of the information obtained by follow-up letters, none the less, this study furnished interesting and thought-provoking data. Although these data do not give conclusive information about the risk of nonoperative and operative treatment of the silent gallstone,

they are noteworthy on this basis: that the patient with silent gallstone may be told that he has about an even chance that symptoms will develop, that he has about one chance out of five that painful seizures will develop and a small chance that jaundice will develop within ten to 20 years. In addition, he should be told that the risk of surgical intervention at the best is about 0.5 per cent when cholecystectomy is performed before complications develop, that the risk will increase to about 3 per cent if he defers surgical intervention until complications develop, or old age and physical debilities appear, but that the increase in risk is counterbalanced by the fact that if he defers surgical intervention he may never require operation. It is not possible on the basis of information now available to tell the patient whether the risk is greater or smaller if operation is performed while the gallstones are silent than if it is postponed until symptoms develop. Certainly, the mortality rate will be low regardless of the choice made.

The reaction of the patient to his problem will be an important determining factor. Many patients will prefer to have gallstones removed in order to eliminate the threat of painful seizures or severe complications. Others, knowing the higher risk of surgical intervention should complications develop, will prefer to take the chance that no symptoms will develop. Surgical treatment of the silent gallstone may be classified as optional or elective surgery, but surgical intervention should not be postponed after symptoms, and more especially after attacks of colic, appear.

CONCLUSIONS

Developments over a period of ten to 20 years in 112 cases of silent gallstones have been ascertained. In 61 cases symptoms did not develop. In the 51 cases in which symptoms occurred, 30 patients complained only of dyspepsia and 21 experienced painful seizures. Five of the 21 patients experiencing painful seizures also had jaundice.

Cholecystectomy may be advised but need not be urged, if the patient prefers to accept the chance of experiencing painful seizures or the increased risk of surgical treatment in the event the complication of calculous disease of the biliary tract appears.

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THORACO-ABDOMINAL APPROACH FOR PORTACAVAL ANASTOMOSIS*

WITH A CASE REPORT OF PORTACAVAL SHUNT EMPLOYING THIS METHOD

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THE WORK OF BLAKEMORE and Lord,[1] Whipple,[2] and Blakemore,[3] has established portacaval shunt as an effective means of treating portal hypertension in properly chosen cases. The indication for, the results of, and the problems attending this operation have recently been reviewed by Blakemore.[4]

The usual approach for portal vein shunt operations (portacaval or splenorenal), is through a wide transverse or an extensive left paramedian abdominal incision. By means of pressure readings of the various parts of the portal system, one can determine whether a portacaval or splenorenal anastomosis is indicated. In general, if either one can be used, as is the case in the intrahepatic block of cirrhosis of the liver, the portacaval shunt is the more desirable, since it offers a larger shunt.[5] The difficulty with the portacaval procedure, however, lies in the fact that the exposure is hardly adequate, necessitating working in a small area under the costal cage, in a deep wound. For this reason many surgeons have been employing the less efficacious but easier splenorenal shunt in instances of intrahepatic block.

It occurred to the writer that a transthoracic, thoraco-abdominal approach to the hilum of the liver would not only give more direct access to the region and present the structures of the hilum and inferior vena cava more superficially but would permit freer mobilization of the liver by permitting its dislocation into the right thorax through the incised diaphragm. Moreover, by placing the patient in a true lateral position, with the right side up, the relationship of the common duct and hepatic artery to the portal vein could be altered from the surgeon's point of view; the portal vein would now be lying to one side instead of behind the above-named structures, thereby excluding the necessity of dissecting the common duct and hepatic artery in order to reach the portal vein. Finally, because of the better exposure and the freer mobilization of the tissues in this region, it would be possible to dissect out the portal vein beyond its point of bifurcation; the branches of the portal vein could therefore be tied and a greater length of the portal vein be preserved for its anastomosis to the inferior vena cava. Obviously, the added advantage of the greater mobilization of the portal vein lies in the fact that there is less danger of tension at the suture line. Then, too, by ligating the branches of the portal vein, the stump is reduced to a minimum, thereby reducing the possibility of an ascending thrombosis initiating in a blind venous pouch.

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With these advantages in view, starting on January 17, 1948, six dogs were subjected to the thoraco-abdominal approach for portacaval anastomosis. In four dogs an incision was made along the course of the ninth rib, from the right lateral margin of the erector spinae muscles to the lateral border of the right rectus muscle. In two dogs the thoraco-abdominal approach was made through the eighth interspace. Either method can be used, depending on the individual surgeon's preference for rib resection or intercostal incision. It

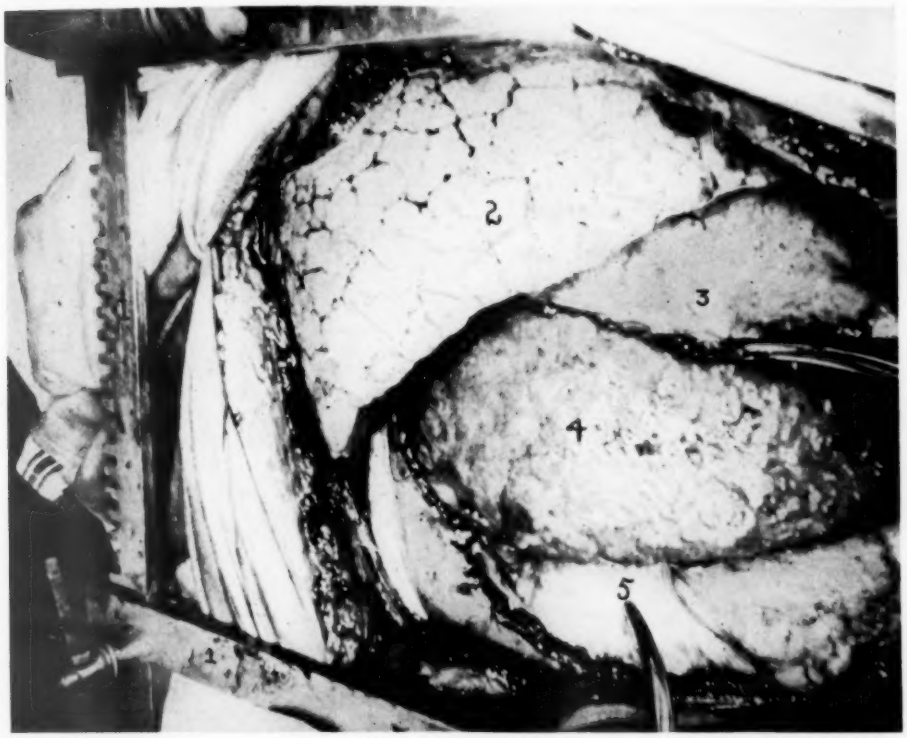

FIG. 1.—The exposure made possible by thoraco-abdominal approach: 1. Finochietto rib spreader between eighth and tenth ribs; 2. Lower lobe of the right lung; 3. Upper leaf of incised diaphragm; 4. Liver. Note marked cirrhosis; 5. Clamp points to lower leaf of severed diaphragm. The wound extends well to the right of area shown in this picture.

may be advisable in some instances to resect the eighth rib, depending on the size of the liver. In each dog the costal cage was opened under positive pressure anesthesia and the diaphragm incised along the line of the wound in the direction of its fibers. The incision was extended through the abdominal muscles to the lateral border of the right rectus muscle, and the peritoneum in this region was opened so as to unite the right hemi-thorax with the abdominal cavity. The intestines were packed away, and the hilum of the liver, the right kidney, the right adrenal gland and the inferior vena cava were easily exposed. The peritoneum over the inferior vena cava was incised and the inferior vena cava was mobilized from the renal vein to the point where it

passed under the liver. The portal vein was then approached; it was clearly discernible to the lateral side of the common duct and hepatic artery. The portal vein was mobilized along its entire length, up to the point where it enters the substance of the liver after branching. It was ligated beyond the point of bifurcation and transfixed by suture-ligature just at the point of bifurcation. The lower portion of the portal vein was compressed by means of a Blalock clamp. A clamp with a deep curve was then placed over the

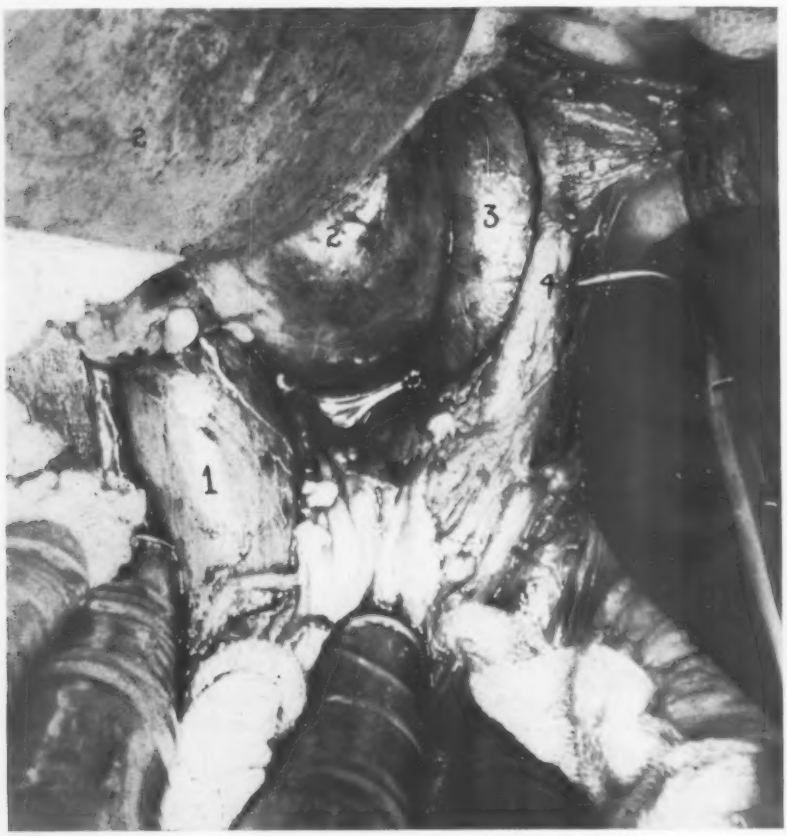

FIG. 2.—Following retraction, the structures to be dealt with are exposed: 1. Inferior vena cava. Note strands of adventitia not yet dissected away; 2. Liver; 3. Portal vein; 4. Common duct pointed out by clamp.

right superior aspect of the inferior vena cava above the renal vein so that the passage of blood would not be obstructed during the anastomosis. A piece of vein between the clamp was excised so as to prevent closure of the aperture postoperatively. A direct end-to-side anastomosis between the portal vein and the inferior vena cava was successfully carried out in all six dogs by use of an everting suture, as described by Blalock,[6] thereby effecting an intima to intima approximation of the vessels. No. ooooo braided black silk on an atraumatic needle was used.

On January 28, 1948, this technic was employed on a 45-year-old male patient suffering from cirrhosis of the liver; he had experienced four episodes of nearly exsanguinating hemorrhage from rupture of esophageal varices, and at the time of operation exhibited a moderate degree of ascites. The operation

FIG. 3.—Completed posterior layer of anastomosis: 1. Stilles rubber-shod kidney clamp on inferior vena cava; 2. Blalock clamp; 3. Portal vein; 4. A forceps holding away anterior layer of portal vein. Note the intima-to-intima anastomosis of these vessels.

was performed under intratracheal cyclopropane anesthesia, with the patient lying on his left side in the true lateral position, supported by a sand bag under the left loin. The ninth rib was resected subperiosteally from its angle to the costochondral junction. The costochondral junction at this point was

cut through, connecting the right thorax with the abdomen. The diaphragm and the muscles were incised along the line of incision. Bleeders from the edges of the diaphragm were clamped and ligated. The intestines were packed away and, because of the posterior lateral exposure afforded by this approach, the need for resection of adhesions from a previous anterior abdominal operation was completely obviated. An excellent view of the hilum of the liver, the right kidney, and inferior vena cava was obtained. The peritoneum lateral to the inferior vena cava was incised. At this time the right adrenal gland was brought clearly into the field. The inferior vena cava was mobilized and vaselinized umbilical tape was passed around it for better control of the vessel. Beginning laterally, so as to avoid injury to the common duct, the portal vein was then mobilized from its origin to beyond the point of its bifurcation. It was possible easily to mobilize the common duct along its entire length, suggesting the value of this approach for difficult hilar reparative surgery of the common duct. The hepatic artery did not come into view during the entire procedure. By means of a Lahey clamp, the right and left branches of the portal vein were dissected out, and double No. 24 cotton ligatures were passed around each. Vaselinized umbilical tape was placed around the lower region of the portal vein, and a small tributary to the portal vein near the hilum of the liver was ligated and cut. The portal vein was then placed between the jaws of a Blalock clamp. At this time an attempt was made to take pressure readings of the portal vein but unfortunately the manometer system did not work satisfactorily. The Blalock clamp was then closed and the sutures which had been placed around the branches of the portal vein were tied. A transfixion suture-ligature of No. 24 cotton was then placed in the portal vein at the point of bifurcation. The portal vein was cut transversely between the ligatures and the Blalock clamp which had been placed well away from the transfixion ligature. By using the handle of the clamp for traction, mobilization of the portal vein circumferentially was easily performed down to the origin of the vessel. A Stille's rubber-shod kidney clamp was then placed on the inferior vena cava above the renals, as described in the discussion of the animal work.*

An incision was made longitudinally into the inferior vena cava for about 3 cm., corresponding to the width of the severed portal vein. A portion of the inferior vena cava was excised along the line of incision (approximately 2 mm. in the center and 1 mm. at the ends),* so as to preclude the possibility of postoperative spontaneous closure. The site selected for anastomosis insured against any possibility of angulation. A direct-end-to-side intima-to-intima anastomosis between the portal vein and inferior vena cava was then established by means of a running everting mattress suture. The suture was tied on the outside at one end and continued across the posterior layer to the opposite margin where a knot was again tied on the outside. These knots prevent pursestringing of the suture line. The anterior layer was likewise

* A special vena cava clamp and a curved long-handled scissor for excising a portion of the vein have since been devised. (They are manufactured by George P. Pilling and Son Company, Phila., Pa.)

FIG. 4.—Completed anterior layer of anastomosis: 1. Liver; 2. Blalock clamp; 3. Portal vein; 4. Suture line; 5. Kidney clamp on inferior vena cava.

closed with an everting running mattress suture. A single tension suture was placed at each side of the anastomosis and one of the free ends of these sutures was tied to a free end of the anastomatic sutures. During the performance of the anastomosis, the accessible intima of both the inferior vena cava and portal veins were repeatedly irrigated with a heparin-saline mixture (one-half cc. heparin to a medicine glass full of saline), as suggested by Blakemore.[7] With the good exposure afforded by this approach, the curve of the needle on the No. 00000 braided black silk proved to be most satisfactory.

FIG. 5.—Anastomosis shown after removal of clamps: 1. Liver; 2. Portal vein; 3. Suture line; 4. Inferior vena cava; 5. Clamp points to right renal vein entering inferior vena cava; 6. Right adrenal gland lies in this area.

After the completion of the anastomosis the Blalock clamp was released. There was slight leakage through the small apertures in the vessels made by the passage of the needle. In a few moments, however, the suture line became perfectly dry. The kidney clamp was then removed, and blood was seen to course through the portal vein up into the inferior vena cava very rapidly. The shunt was more than adequate, measuring approximately 2½ cm. in

diameter, and the strength of the one-layer suture line was put to visible test by the fact that with the patient lying on his left side maximum pull was exerted on the anastomosis by the weight of the intestines. A mixture of plasma and thrombin was injected into the region of the anastomosis. The rent in the posterior peritoneum was sutured. Fifty thousand units of penicillin was placed in the peritoneal cavity and another 50,000 units in the chest. The peritoneal cavity was closed. The diaphragm was repaired with interrupted imbricating mattress sutures, and the chest and abdomen were closed in layers. An intercostal drainage tube below the line of incision was placed in the chest and connected with a water-sealed drainage system. Cotton suture technic was employed throughout. Actual operative time was approximately four hours. The author feels quite sure, however, that in the future the portacaval shunt can be performed in well under three hours by means of this approach.

The severe portal hypertension that existed in this patient and the great speed with which the blood rushed through the anastomosis, coupled with the precaution of having excised a portion of the inferior vena cava before commencing the anastomosis gave assurance that the shunt would not close off. and argued against the need for postoperative parenteral administration of heparin.

Three pints of blood were used during the operation. Postoperatively, the patient was placed in the Trendelenburg position so as to prevent gravity drag on the portal vein; this was maintained for 48 hours, at which time it was felt that sufficient adhesion formation had fixed the portacaval anastomosis and the adjacent tissues in a favorable, tensionless position, and obviated the possibility of postoperative angulation due to variations in posture. The patient received parenteral fluids for three days and was permitted out of bed on the fourth postoperative day. A detailed account of the medical aspects of this case and the results obtained by this procedure will be reported in a future communication.

DISCUSSION

A direct anatomic approach to the hilar structures of the liver seemed a logical and necessary step if surgeons hope to advance and popularize surgical procedures on these structures. The writer felt, for instance, that portacaval anastomosis, physiologically valuable in the treatment of portal hypertension. was not taking its proper place because of a technically difficult and unbelievingly time-consuming approach requiring from 7 to 12 hours by the abdominal route. The average physician hesitates to entrust a bad risk patient to the average surgeon for so traumatic a procedure. With the above described approach, however, portacaval anastomosis becomes a relatively simple operation, carrying no more technical difficulty than already mastered abdominal and thoracic operations.

An objection to the thoraco-abdominal approach may be raised by those who feel that it is imperative to obtain pressure readings of the portal system at the time of operation, and that a surgeon should be free to make a choice

between portacaval and splenorenal anastomosis. In most instances, a diagnosis usually can be made preoperatively or at a previous operation. However, the objections would be well taken were it not for the fact that in the thoraco-abdominal approach the abdominal portion of the incision could be made first, and pressure readings taken. If a splenorenal anastomosis is decided upon, the incision could be enlarged to the left side so as to permit adequate exposure for this procedure. If, on the other hand, a free choice exists, then by all means the larger portacaval shunt is desirable and could be performed more easily than the splenorenal shunt, by extending the incision over the costal cage. By means of the thoracoabdominal approach the portacaval anastomosis is certainly easier to perform than a splenorenal shunt, where often exsanguinating hemorrhage is encountered in dealing with an enlarged spleen complicated by perisplenitis and extensive collateral circulation. Moreover, the thoracoabdominal approach reduces to an absolute minimum the handling of bowel, and thereby avoids troublesome ileus.

Because of the excellent exposure of the entire length of the common duct, the approach described is strongly urged for reparative common duct surgery where reconstruction work must be carried out close to the hilum of the liver. It will also prove most useful for easy and complete exposure of the right adrenal and should result in a wider scope of activity in dealing with conditions involving this gland.

SUMMARY

A new approach for a portacaval anastomosis, common duct surgery, and right adrenal gland surgery has been described. The advantages of this transthoracic, thoraco-abdominal exposure are as follows:

1. The structures in the hepato-gastric omentum are approached from the lateral aspect, thereby presenting the portal vein to the side of the common duct and the hepatic artery.

2. The liver can be dislocated into the right hemi-thorax, since the diaphragm no longer restricts mobilization of the liver as it does in the abdominal approach.

3. Access to the hilum of the liver is simplified, enabling ligation of the right and left branches of the portal vein instead of the portal vein itself. High ligation not only affords a longer segment of portal vein for anastomosis but reduces the chances for thrombosis formation in a blind venous pouch.

4. Exposure is wide because of the free spread of the ribs and the free communication of the chest and abdominal cavities.

5. The liver hilum structures and the inferior vena cava are presented more superficially than when the patient is placed on his back.

6. With the patient lying on his left side the suture line supports its maximum tension, and dependability may be visualized at the time of operation.

7. Handling of the intestines is avoided, thereby reducing to a minimum postoperative ileus.

8. The troublesome venous collateral circulation is easily coped with by this direct exposure; more importantly, it is practically avoided.

9. By employing the thoraco-abdominal route, portacaval shunts will be favored over the less desirable splenorenal junctures, and will make for a less hazardous postoperative course since the use of heparin is scarcely called for with the larger portacaval anastomosis.

10. Pressure reading of the portal system can be taken at the time of operation simply by performing the abdominal portion of the thoraco-abdominal wound first.

11. Little chance for postoperative incisional hernia exists, since the abdominal portion of the wound constitutes a small part of the entire incision.

12. Excellent exposure is offered by this route both for reconstructive surgery of the common duct where the injury is close to the hilum of the liver and for right adrenal gland surgery.

CONCLUSIONS

The thoracoabdominal approach as described by the author and as successfully performed by him on a 45-year-old male patient with cirrhosis of the liver appears to be the procedure of choice in dealing with portacaval anastomosis, reconstruction of the common duct close to the hilum of the liver, and surgery of the right adrenal gland.

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The photographs presented in this article were taken by Dr. Charles P. Bailey and Miss Mildred Updite, R.N.

THE RIGHT THORACO-ABDOMINAL APPROACH*

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THE NEED FOR AN APPROACH to the contents of the right upper quadrant of the abdomen which would provide more satisfactory exposure, has long been appreciated. This has been especially true in regard to patients with biliary tract disease who are, as a rule, quite obese.

Numerous surgeons, including one of us (J. P. H.) had occasion during the war to operate upon patients with wounds involving both the thorax and abdomen which required exploration of both cavities, on the right side as well as on the left. Thus far, we have not heard of the elective use of an approach to the right upper quadrant via a segmental thoraco-abdominal incision, although Carter has speculated about the possibilities of such an incision for lesions of the right lobe of the liver.

For an appreciable period, we have had under consideration the right thoraco-abdominal approach which, it was believed, might offer the solution sought to the problem of exposure. At first, it seemed apparent that the right lobe of the liver would certainly become more readily accessible for safe surgical attack, not only because of the exposure of the lobe *per se* but because the inferior vena cava might also be reached and temporarily controlled during the process of excision, *above* the liver as well as below.

Further, it occurred to us that in procedures upon the biliary tract through the conventional abdominal route, it was primarily the immobility of the costal margin which prevented good exposure. By using the segmental thoraco-abdominal incision, the costal margin might thus be eliminated as a barrier. The right lobe of the liver could then be gently rotated backward and upward into the lower thorax through a radial diaphragmatic incision. As a result, the biliary tract and porta hepatis, which usually nestle deep in the posterior part of the right upper quadrant, overhung by the liver, would be exposed easily and consistently in a manner heretofore achieved only occasionally and then, as a rule, in thin patients.

With these ideas in mind, the incision was first used in the anatomy laboratory. Through it, a complete dissection of the extrahepatic biliary tract, caval, and pancreatico-duodenal regions was performed. Although the tissues were fixed and the liver lobe rigid, we were encouraged to believe that this approach might indeed be worthwhile.

The maneuver was next tried in dogs. Because of the lobulated nature of the canine liver, it was necessary to reserve judgment as regards the mobility of the liver in man, but these animals served to illustrate that if the

* Submitted for publication, May 1948.

liver could be readily retracted as believed, then certainly the anatomic parts to be dealt with would be adequately uncovered.

The last phase of the preliminary work was done in the postmortem room where the tissues, when relatively fresh, are as nearly of the same consistency as in the living patient. This work demonstrated clearly that the procedure deserved clinical trial.

It was decided that the first case should be a simple cholecystectomy in order that if the patient presented any complication not foreseen by the pre-

FIG. 1.—The patient is placed on a horizontal table with sand bags beneath the right shoulder and buttock and with the abdomino-thoracic region centered over the "break." The table should be of the type which permits rotation in its long axis. This movement facilitates access to any part of the wound.

liminary work, it might still be feasible to remove the organ through the abdominal segment of the incision in a more nearly conventional manner. However, the most obese patient available was selected in order to put the presumed advantages of the procedure to real test.

CASE REPORT

On May 7, 1947, a 45-year-old, Para IV, Puerto Rican housewife presented herself because of severe right upper quadrant pain of 24 hours' duration. For 14 years she had suffered from intolerance to fatty foods with indigestion and belching. This was sometimes accompanied by right upper quadrant and epigastric discomfort which infrequently radiated through to the scapular region. During attacks, she occasionally had nausea and vomiting but had never had chills, fever or jaundice. The episode which brought her to the hospital seemed more severe than previous ones. She vomited more intensely and felt "chilly" but did not record her temperature. The pain radiated to the

lower angle of the right scapula and to the right shoulder. She had noted that her urine was darker than usual, but two stools passed after onset of her present illness were normal in color. She had never had typhoid fever. Past history and inventory of systems were otherwise non-contributory.

Physical Examination.—Temperature 100.2; Pulse 80; Weight 195 lbs.; Height 61."

The patient was a well developed, very obese woman in no distress. The skin and sclera were clear. The abdomen was protuberant with a very thick abdominal wall. There was moderate spasm in the right upper quadrant with direct and rebound tenderness in that area. No mass or viscus was palpable. The remainder of the physical examination revealed no further relevant abnormal findings.

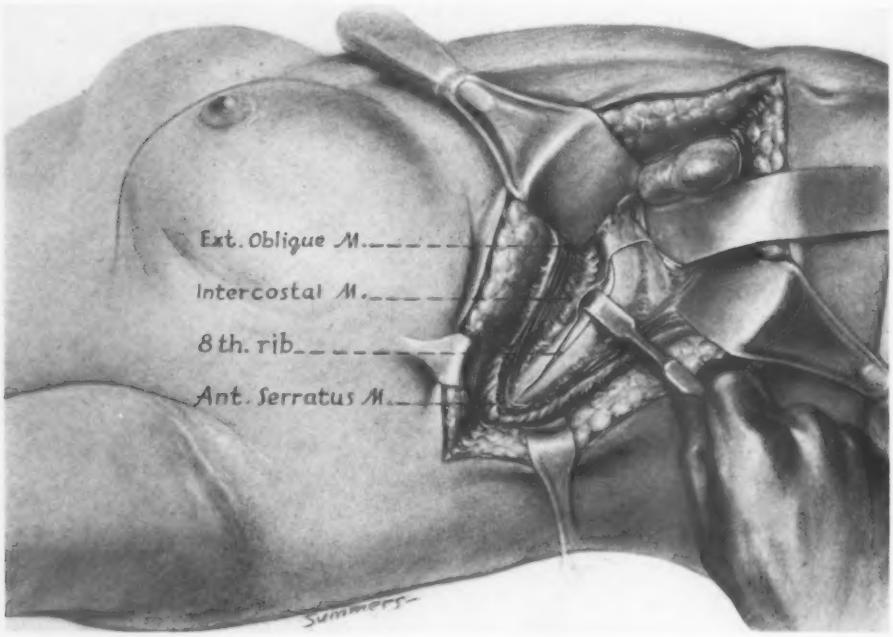

FIG. 2.—The abdomen has been entered; an exploration can be made through this segment of the wound. The 8th rib is then resected subperiosteally after which the costal margin is divided. Some operators might prefer an intercostal approach.

The impression at the time of admission was:

1. Acute cholecystitis superimposed on chronic cholecystitis with cholelithiasis.
2. Obesity.

Laboratory Findings.—Studies following her admission revealed a serum bilirubin of 3.6; alkaline phosphase 6.4; white blood count 10,100 with 53 per cent polymorphonuclears; the urinalysis showed four plus bile. An electrocardiogram was negative as was a chest film. On symptomatic care and a low fat, low caloric diet, the patient improved. A cholecystogram on the 11th hospital day revealed the presence of dye in the gallbladder with a 3 cm. area of translucency, which was interpreted as a single large stone. She was, therefore, prepared for cholecystectomy and operated on the 14th hospital day.

Pathologic Findings.—The gallbladder was moderately distended and contained several large facetted stones, one of which was tightly impacted in the ampulla. The cystic duct was thickened but the common duct showed no evidence of disease. The duodenum, head of the pancreas, right lobe of the liver, and stomach presented no abnormalities.

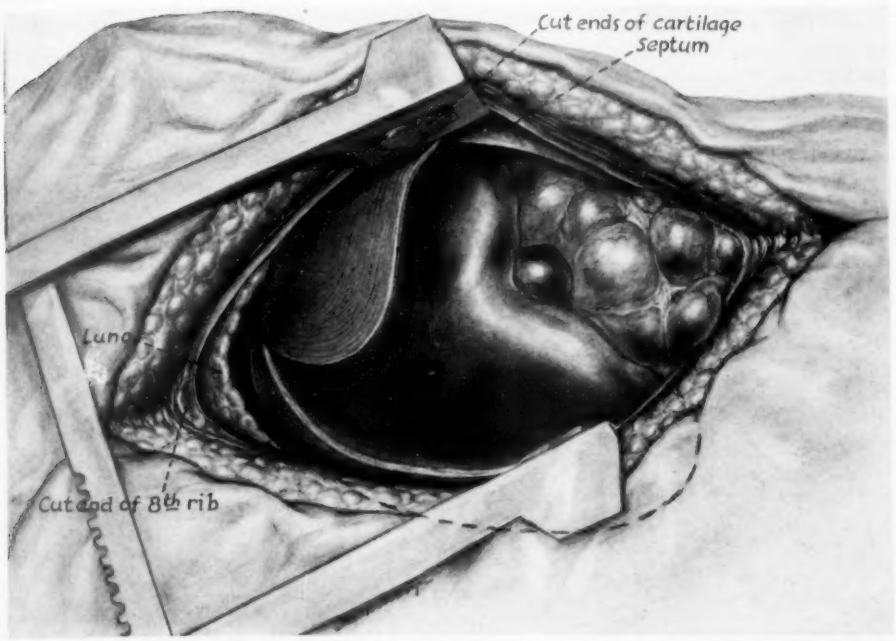

FIG. 3.—The addition of the radial incision to the diaphragm, from costal margin to dome, reveals the antero-superior surface of the liver. The lower lobe of the lung is evident above; the gallbladder, fundus and colon appear below. Note the cut edges of the diaphragm which have been separated, along with the mural components of the incision, by the Finochetto retractor.

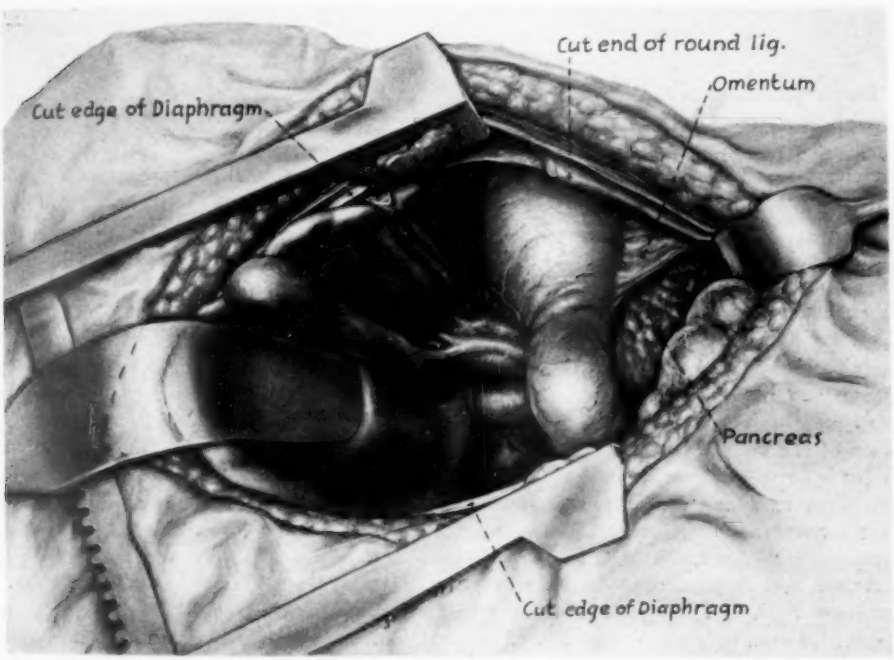

FIG. 4.—The right lobe of the liver has been turned upward through the diaphragmatic defect into the lower chest. The structures formerly overhung by the liver, are thereby uncovered.

Operative Procedure. On May 22, 1947, the patient was anesthetized with a combination of agents administered intratracheally. A long curved incision was made overlying the ninth interspace. It extended from the mid-axillary line downward and forward across the costal margin, and over the abdomen to the linea alba at a point 5 cm. above the umbilicus. This incision was carried through the subcutaneous tissue to expose the external oblique muscle and anterior rectus sheath. The latter was divided transversely as was the right rectus muscle. The external oblique muscle was split in the direction of its fibers, whereas the internal oblique, and transversus abdominis muscles were

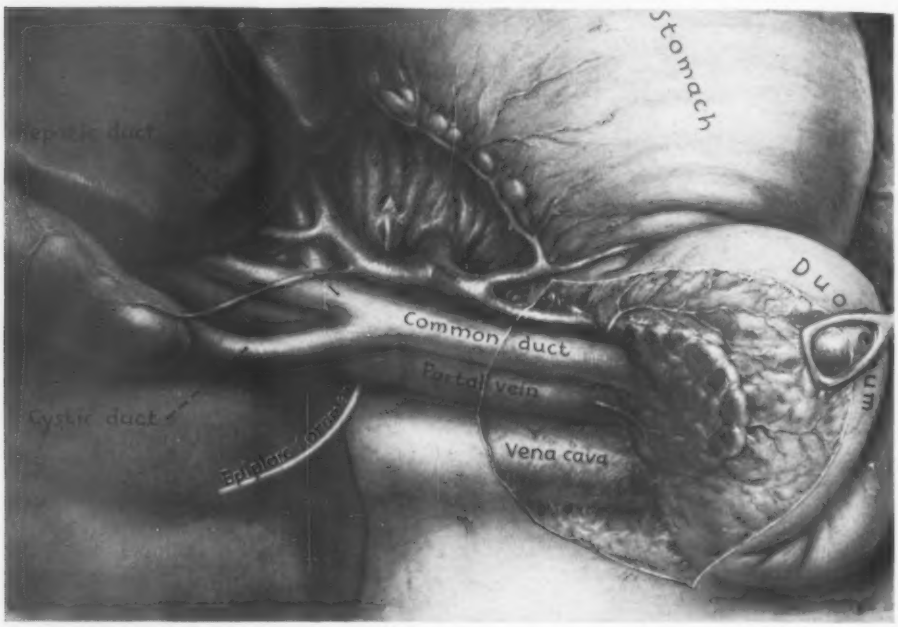

FIG. 5.—This enlargement, which shows the duodenum reflected, was made from a dissection in the postmortem room. It illustrates the extensive exposure of the biliary tree and allied structures, which is possible through this approach.

transected in the line of the incision. The common costal cartilage was divided between the 9th and 10th ribs and the chest entered through the 9th interspace. The abdomen was then entered by means of a radial incision in the diaphragm which extended from its periphery at the point of division of the costal margin, to the right dome. Following this the peritoneum was divided throughout the length of the abdominal segment of the wound. A Finochietto retractor was placed between the 9th and 10th ribs and spread. In this manner, the wound was held wide open without other retraction. Next, the right lobe of the liver was gently rotated upward through the diaphragmatic defect into the right lower chest, exposing the gallbladder, the porta hepatis, the gastrohepatic omentum, the vena cava and the pancreatico-duodenal region admirably, in spite of the patient's great obesity. The stomach and hepatic flexure were packed off with moist abdominal pads; several filmy adhesions between the duodenum and gallbladder were divided. The common and cystic ducts were identified and isolated as were the common hepatic duct, and the right and left hepatic ducts with their accompanying vessels. This dissection permitted unusually extensive study of the duct system. The cystic duct was then divided between clamps. The proximal end was ligated with a transfixion suture of No. 000 Deknatel. The cystic artery was also divided and ligated with the same material. The

gallbladder was dissected from the liver, from ampulla to fundus, without difficulty after which the bed was repaired with interrupted silk sutures. The peritoneum over the cystic duct stump and in the region of the porta hepatis was also approximated by means of interrupted sutures. After the liver had been replaced in its normal position, a cigarette drain was inserted into Morrison's pouch and brought out along the gallbladder bed through the abdominal part of the incision. The diaphragm was then closed with interrupted No. 000 Deknatel. A single suture of No. 0 Deknatel was used to approximate the cut edges of the costal margin. The thoracic and abdominal mural components of the incision were then closed anatomically with interrupted sutures of No. 000 silk. The patient's condition during and after operation was excellent.

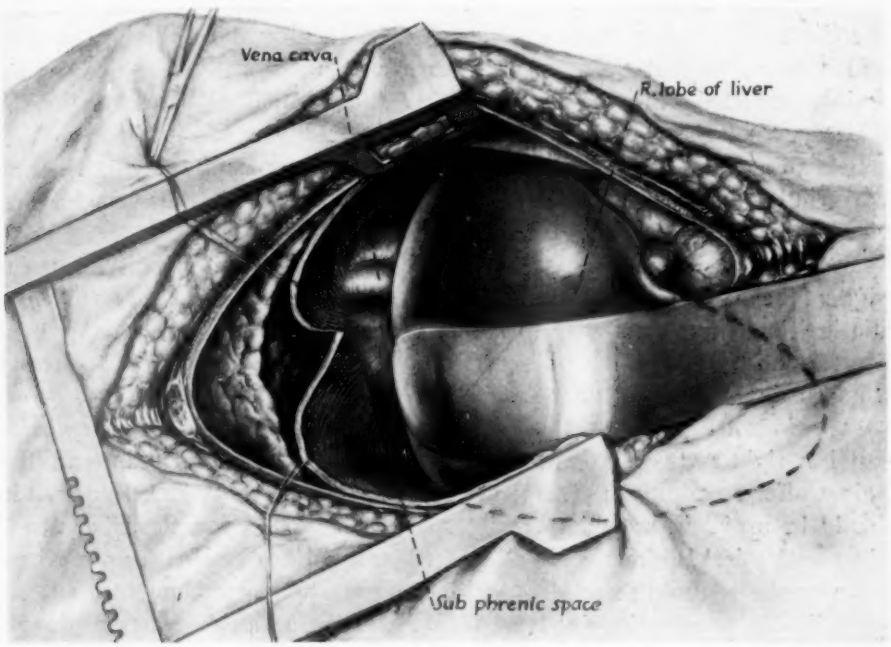

FIG. 6.—The liver has been retracted downward; the diaphragm edges are suspended by means of silk sutures to show the position of the inferior vena cava *above* the liver. The liver lobe is also outlined to demonstrate its position. (See Text.)

Postoperatively, the temperature rose to 100.2 on the first day; 102.6 on the second; 101.8 on the third and then declined gradually to normal. This fever was due in part to transient atelectasis of the right lower lobe, which responded to coughing efforts. Her convalescence and wound healing were otherwise favorable and uneventful. She was discharged on the 15th postoperative day. She has been followed in the outpatient clinic since operation and has remained well.

DISCUSSION

This cholecystectomy was performed with ease in spite of the patient's unusual obesity. There was no tugging upon retractors and no difficulty in exposure. The right lobe of the liver was easily rotated upward into the lower thorax and produced no change in the patient's general well being. There was no torsion of the inferior vena cava or structures of the hepatic

trinity. The cystic duct and artery were dealt with under direct vision in a wide open wound, rather than in one overhung by the liver, costal margin and abdominal wall. Not only was the common hepatic duct readily accessible throughout its entire extent, but each of the right and left hepatic ducts was accessible over a distance of 1½ to 2 cms., permitting unusual certainty in palpating for stones or, had it been necessary, the performance of high biliary-intestinal anastomosis.

The phase of the operation which was potentially the most hazardous, namely, the division of the cystic artery and duct, was rendered less so because of the exposure afforded by the maneuver of displacing the liver into the chest. This latter act is accomplished by turning the right lobe upward and outward so that the inferior surface, which originally faced downward and posteriorly, comes to face anteriorly and still slightly downward. The liver does not rotate as if upon a hinge but readily molds into the position sought in a "fluid" type of movement. During the process, the portal fissure opens in the same manner as a single pleat of an accordion, thus bringing into view a greater extent of the biliary tree.

Our particular patient brought out only one unforeseen difficulty. The cut edges of the diaphragm retracted so that at the peripheral part of the diaphragmatic incision, it was not easy to suture the edge because of the friable liver beneath and the unyielding rib superficially. It is probable that a rib resection would make closure less difficult.

We do not believe that this approach should supplant the ordinary paracostal or right rectus incision for cholecystectomy in the average instance but it does offer an additional degree of exposure for exceedingly obese cases. It should be useful in secondary and tertiary operations upon the biliary tree, when anatomical landmarks have been obliterated by adhesions from previous procedures. It would seem ideal for plastic operations upon the duct system. Secondly, the pancreatico-duodenal region was also superbly exposed, though this latter region was not the one in which we were primarily interested. We have witnessed numbers of pancreatico-duodenectomies through both transverse and vertical abdominal incisions in which that operative field was less well exposed than in this case. Thirdly, the portal vein and inferior vena cava are readily evident throughout much of their proximal extent; as a result the incision should be useful in performing portocaval anastomosis.* Fourthly, the right lobe of the liver is exceptionally well exposed and its removal might be made safer by the temporary control of the inferior vena cava *above* the liver, as well as below. Fifthly, in selected patients who have an unusually high diaphragm, in addition to their other indications for the approach, it is possible that the contents of the right upper quadrant might be satisfactorily exposed through the chest and diaphragm alone, without the addition of the abdominal mural segment of the incision. Thus the gallbladder might be more

* This approach has recently been used by Dr. Arthur Blakemore in a case of portal hypertension. Portocaval anastomosis was accomplished with excellent exposure. Postoperative recovery was uncomplicated.

easily removed in some cases through the chest than through the abdomen, just as splenectomy and gastrectomy are sometimes more satisfactorily accomplished from *above*, on the left side of the abdomen. The incision should also be valuable in the occasional inflammatory or neoplastic lesion which traverses the diaphragm to involve both the right lower lobe of the lung and the right lobe of the liver.

Lastly, if one encounters unusual difficulty while operating upon right upper quadrant structures through a conventional abdominal incision, the segmental mural and diaphragmatic components may be added to obtain the additional exposure which is so valuable in time of need. This combination of ordinary abdominal incision, plus subsequent addition of the segmental components, has been used to advantage on the left in numbers of cases, without compromise of circulation of the flaps so formed.

CONCLUSION

We believe that the right thoraco-abdominal segmental approach offers valuable additional exposure in properly selected operations upon the biliary tree, the pancreatico-duodenal region, the portocaval region and the right lobe of the liver.

If unexpected technical difficulties are encountered in the right upper quadrant and additional exposure would be advantageous, it can be used as an adjunct to the ordinary right rectus, paracostal, or transverse abdominal incisions.

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EXPERIENCE WITH THREE THOUSAND CASES OF BRACHIAL PLEXUS BLOCK; ITS DANGERS

Report of a Fatal Case

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FROM THE ORTHOPEDIC SERVICE OF DR. GONZALEZ-AGUILAR, CASA DE SALUD "VALDECILLA," SANTANDER, SPAIN

IN JULY OF 1936 we published a paper dealing with 250 cases of Block Anesthesia of the Brachial Plexus.[20] We expressed at that time our surprise at the lack of interest which the majority of surgeons manifest in this procedure. Moreover, we pointed out that a definite dearth of literature existed concerning brachial plexus block. Shortly after our report in July of 1936 a thesis for doctorate[15] was published dealing with 403 cases of block anesthesia of the brachial plexus.

Owing to the great number of war casualties with wounds of the upper extremities which came under our care at the Orthopedic Services of the Casa de Salud-Valdecilla and the Hospital Militar Cantabro, we have been able to compile 3000 cases in which block anesthesia of the brachial plexus has been used. In the present paper we shall not discuss fundamental considerations of this procedure, nor anatomic details and technics. Instead, drawing from our experience, we shall confine ourselves to a discussion of the accidents encountered in the use of brachial plexus block, one of which in our series proved fatal. We hope, also, to elucidate the causes of such accidents; thus aiding in their prevention and raising the procedure of brachial plexus block to a more deserved place in the surgery of the upper extremity.

In practically all of our cases, the technic described by Kulenkampff,[14] and expanded by us in our previous paper, was employed.

The supraclavicular region contains important anatomic structures with which we must be completely familiar if we would obviate accidents. It is situated over the dome of the pleura and is traversed by major nerves and blood vessels. This region is also noted for the rapidity with which drugs are absorbed by it; hence the dosage and toxicity of the anesthetic agent should be carefully borne in mind. With the foregoing facts as a basis, the accidents encountered in brachial plexus block are classifiable into pleuropulmonary, neural and vascular. However, before discussing these accidents, we shall point out the importance of choosing the proper anesthetic and its dosage. At present we are using novocain "Bayer," a 2 per cent solution "without adrenalin," and in none of our cases have we observed lipotemias or syncopes; only once did a mild degree of cerebral excitation, as is seen in the first stage of ether anesthesia, occur, when the dose exceeded the currently used dose of 20 cc., or when the latter dose was employed in children. Reducing the dose to a suitable amount, we have succeeded in obtaining adequate anesthesia in babies as young as one and one-half years of age. On the other hand, when

constrained by circumstances to employ substitutes for novocain, such as, sincain, sedocain, alocain, etc., it was not uncommon to observe untoward effects of tachycardia, chills, bouts of sweating, and angina pectoris. In one case, when by error novocain with adrenalin was used, tachycardia and

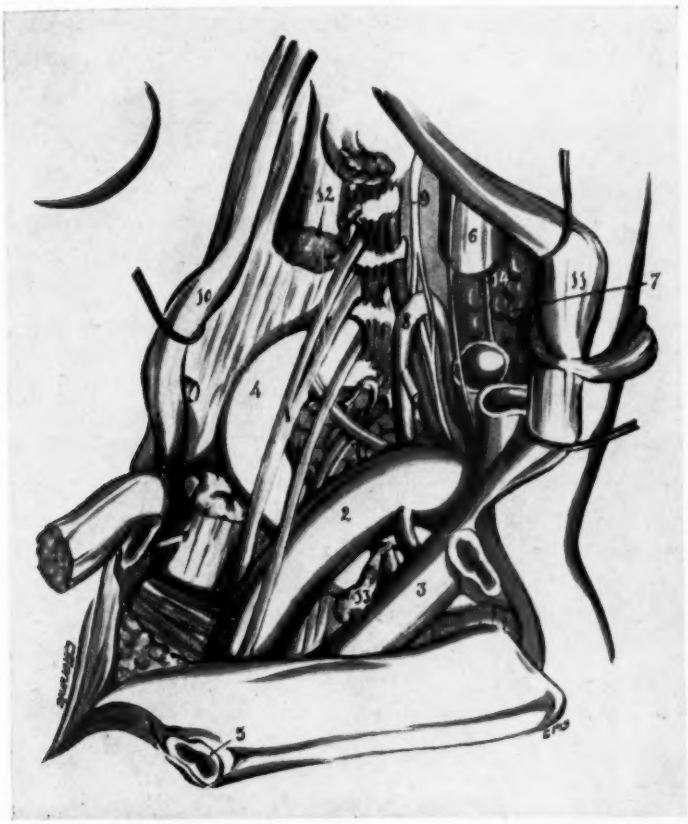

FIG. 1.—Right supraclavicular region, head being turned to the opposite side; the sterno cleido mastoid and trapezius muscles are drawn aside by means of retractors; the omohyoid and the clavicle are sectioned and the latter is pulled down. (1) brachial plexus, (2) subclavian artery, (3) subclavian vein, (4) 1st rib, (5) clavicle, (6) vasculo-nervous plexus of the neck, (7) thyroid, (8) inferior hyoid artery, (9) chain of sympathetic ganglia, (10) trapezius, (11) sternocleidomastoid muscle, (12) posterior scalenus, (13) anterior scalenus, (14) recurrent nerve.

arrhythmia resulted of such a degree, that, though the patient did not expire, death seemed imminent.

To obtain a prolonged anesthesia, we have never employed percain; in all our cases an anesthesia of two hours, produced with novocain, being sufficient. Concentrations of novocain of less than 2 per cent produce poor results, and we deem greater than 2 per cent to be dangerous.

To obviate the untoward effects resulting from both the toxic action of

the drug and from its rapid absorption in the cervical region, we believe it a prudent measure to inject the drug very slowly and to record the rate of the radial pulse.

In the foregoing accidents Coramine has been found efficacious.

Pleuropulmonary Accidents. From a careful review of the literature on brachial plexus block, we have gleaned three reports of death following this procedure; namely, Capelle in 1916,[4] Vischer in 1918,[22] and Hering in 1920.[7]

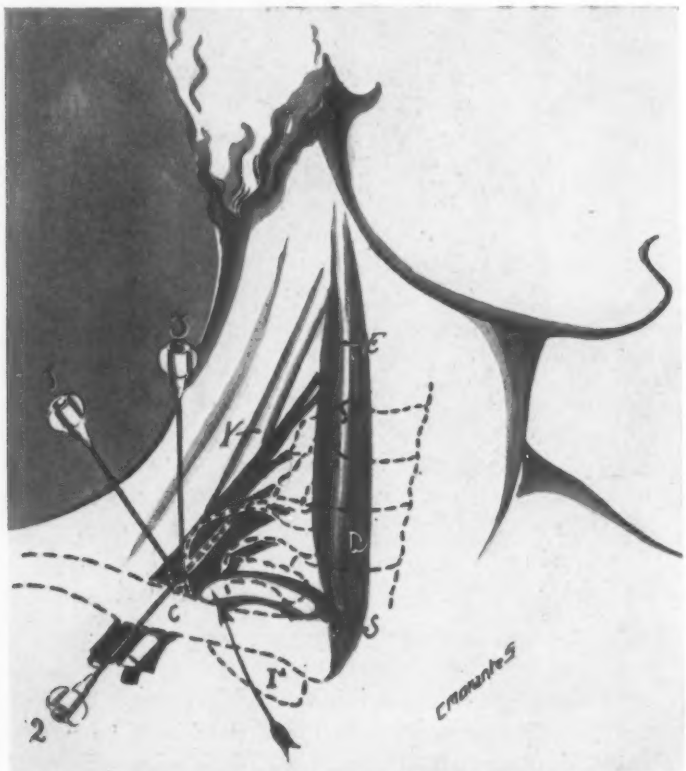

FIG. 2.—Technic of Kulenkampff. (1) 1st stage, (2) 2nd stage, (3) 3rd stage. The arrow indicates the point where one should place the index finger to protect the subclavian artery. (S) subclavian artery; (C) clavicle; (E) anterior border of the trapezius; (Y) posterior border of the sternocleidomastoid muscle; 1 (a) 1st rib.

All of these deaths resulted from trauma to the apical pleural and lung parenchyma. The trauma was produced by incorrect insertion of the injecting needle beneath the first rib as a result of respiratory movement. The supraclavicular technic of Kulenkampff was used by all these surgeons. Following such trauma there supervened rapidly a subcutaneous emphysema, pneumothorax, hemothorax and dyspnea, successively; and in Capelle's case the patient died on the second day. An autopsy revealed, in addition to pneumo-

sema. All the cases showed emphysema and cardiovascular insufficiency. Weil[23] reported a case of spontaneous recovery following mediastinal emphysema.

On five occasions we punctured the parietal pleura the sibilant sound produced by the inrushing air, however, placed us on guard, and the needle was withdrawn carefully. Roentgenograms disclosed the presence of moderate

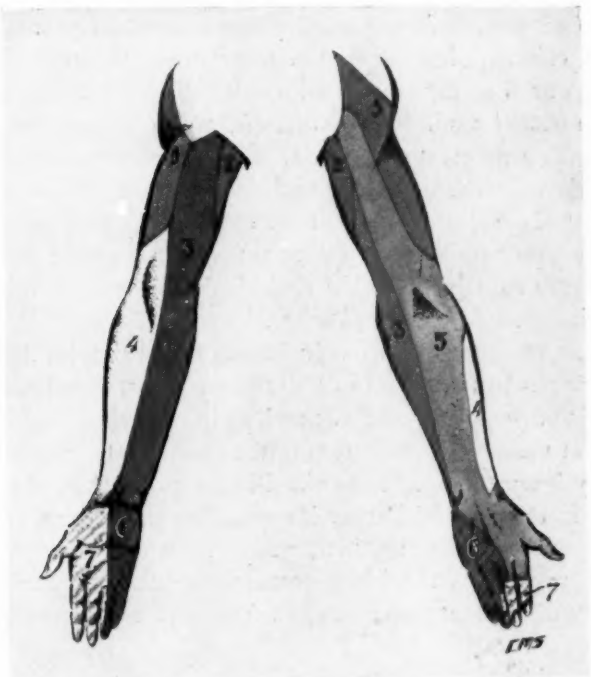

FIG. 3.—Zone of innervation of each of the branches of the brachial plexus. (1) circumflex, (2) accessory of the internal cutaneous brachial nerve and intercostal nerves, (3) internal brachial cutaneous nerve, (4) musculo-cutaneous, (5) radial, (6) cubital, (7) median.

pneumothorax which, together with the consequent dyspnea, subsided in 48 hours.

In order to obviate pleuropulmonary accidents Capelle adopted the axillary route of Hirschel,[8] but as we shall presently point out, this route is no less dangerous than the supraclavicular route. Mulley,[16] in 1919, with the same intention modified the technic of Kulenkampff, placing the point of injection 3 cm. above the clavicle and 2 cm. lateral to the external jugular vein, and maintaining the needle in a more horizontal position. We believe that this modification renders the procedure of brachial plexus block less certain. The same can be said for the infraclavicular technic of Balog,[2] or and of the Anglada Santoni, technic as well as of the Kinn modification.[12]

We consider the paravertebral technic of Kappis[11] to be unusual. To avert pleuropulmonary accidents we maintain that adherence to the technic of Kulenkampff alone is necessary.

Neural Accidents. We shall not consider here the transient aphonias and Claude Bernard-Horner syndromes that supervene from the anesthesia of the recurrent laryngeal nerve and of the cervical sympathetic chain respectively. Hemidiaphragmatic paralysis can occur as a result of phrenic nerve block if excessive infiltration of the region, or incorrect technic is practised. We have, however, on four occasions, produced bilateral brachial plexus block without any untoward effects. Siebres[21] and Klauser[13] report similar successes.

Following our first experience with the technic of Kulenkampff, on one patient there resulted a mild contraction and paresthesia of the lower extremity together with a moderate lipotemia. We conjectured that the needle may have entered the spinal canal through an intervertebral foramen.

Of the neural complications that follow brachial plexus block especially interesting are the meralgias with paresthesias which have been pointed out by many authors; namely, Babizki,[1] Raechke,[19] Flesch-Telesius,[5] Hartler and Keppler,[6] Hirschel,[19] Borchers,[3] Pacher,[17] and Hylkema.[10] These complications all subside, some very slowly. In five of our own cases the complications abated in 20 days. In almost all of these cases a tourniquet was employed to obtain ischemia of the operative field; and, as in the case of Babitzki, meralgias did not occur at the site of application of the tourniquet. In some of the cases, displaced bony fragments may have aided in the production of these complications. Lastly, in the cases of Pacher it seems that the trauma produced by the intraneural injection of the anesthetic will explain these complications.

A thesis[15] previously cited, incorporates the results of a study of the effects of brachial plexus block on the chronaxie. We have never been able to observe any alteration.

Vascular Accidents. Puncture of the common carotid, subclavian, vertebral, and inferior thyroid arteries can, and, in fact, does occur. It has very frequently happened in our own experience. Simple withdrawal of the needle alone is necessary, when it does occur. At times a hematoma will form, but will disappear with the application of a compression bandage. In one of our cases edema of the upper extremity supervened, but subsided after 48 hours with the extremity in the position of abduction.

Intra-arterial injection of the anesthetic produces only a transient state of anesthesia. On the other hand, intravenous injection is dangerous, for the anesthetic is carried to the heart. Moreover, part of the drug may reach the medulla oblongata and produce cardiorespiratory difficulties. Intravenous injection is eight to ten times more toxic than intra-arterial injection, according to Pauchet,[18] even though only one-sixth of the drug ever reaches the cerebrum. We know of no case of death reported as resulting from this mechanism. In the absence of other explanations we believe that perhaps the single case of death in our own series resulted from intravenous injection of the anesthetic.

Report of a Fatal Case. An emaciated woman, age 67, was to be treated for an old fracture of the clavicle at the junction of its middle and inner thirds. The fragments were misplaced and an exuberant callus, the size of an orange, had formed. The patient presented no signs nor symptoms suggestive of pleuropulmonary disease. The supraclavicular technic of Kulenkampff was to be employed. But due to the abnormal anatomic conditions that prevailed, the pulse of the subclavian artery, which is a landmark, could not be felt and a point above the customary midpoint on the clavicle was elected. The needle was directed obliquely to avoid the obstacle presented by the callus, and perhaps in searching for the brachial plexus, the subclavian vein was entered. We suspected that intravenous injection had occurred, when the patient became pallid and cold, and her pulse became weak. The patient lost consciousness, and died in two minutes, despite the

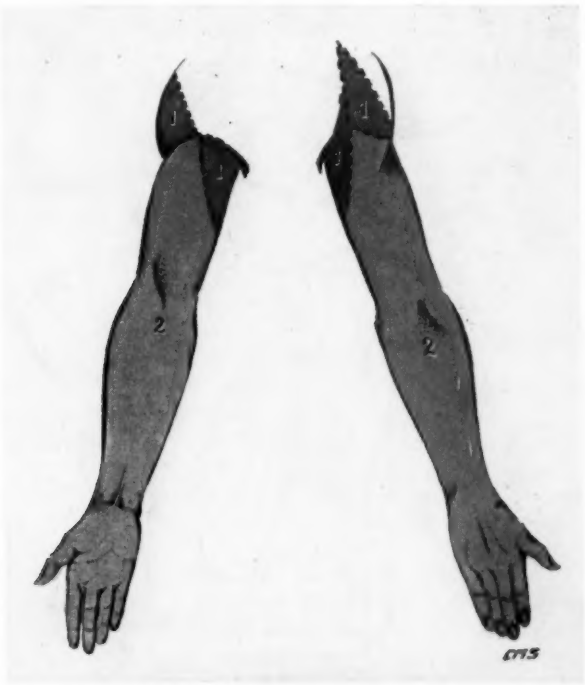

FIG. 4.—Zones of anesthesia of the brachial plexus. (1) Hypoesthesia corresponding to the territories innervated by the branches of the circumflex nerves, 1st and 2nd intercostals. (2) complete anesthesia.

use of cardiovascular tonics and artificial respiration. The anesthetic used was 25 cc. of 2 per cent novocain.

An autopsy performed by Doctor Sanchez-Lucas of the Casa de Salud-Valdecilla, failed to disclose any lesion that would explain this fatality. We believe that the drug entered the venous system through the subclavian vein, was carried to the heart, and subsequently produced its mischief in the central nervous system.

This accident, though fatal, is insufficient to make us abandon this procedure which is so beneficial to the patient and so helpful to the surgeon. In another article we have discussed the advantages of brachial plexus block, and hence we shall not do so here.

Since this fatality, we have successfully employed brachial plexus block in more than 1200 cases. The supraclavicular technic of Kulenkampff was used, and we are favoring this technic more and more each day. We consider the infraclavicular technic of Balog or that of Anglada, Santoni, and above all, the axillary technic of Hirschel to be very dangerous. Our reasons for this statement are that the artery and vein are in proximity in the infraclavicular

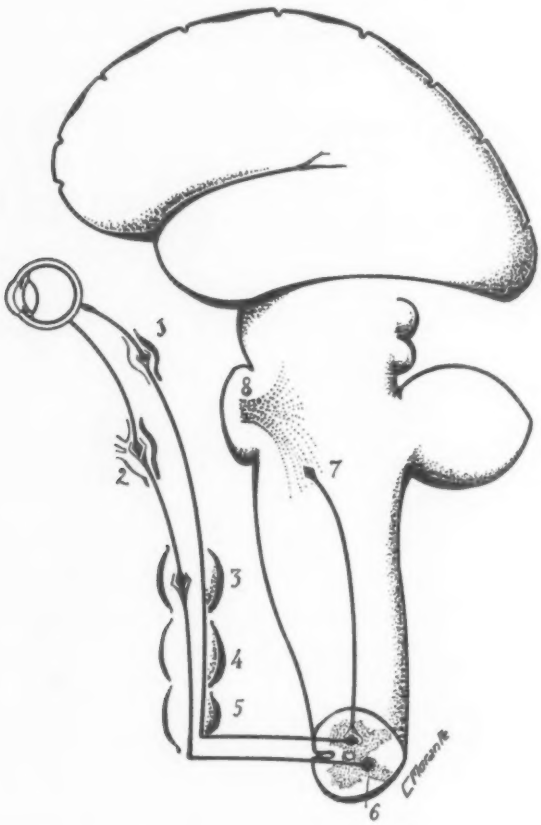

FIG. 5.—Claude Bernard-Horner syndrome. (1) ciliary ganglion, (2) Gasserian ganglion, (3) superior cervical ganglion, (4) median cervical ganglion, (5) inferior cervical ganglion, (6) cilio-spinal center of Budge, (7) Oculo sympathetic bulbar center, (8) descending root of the trigeminus.

region, and that the nerves of the plexus form a network around the vein at the level of the axilla; thus the veins are more easily exposed to accidental puncture.

All of the cases of death and accident, including our own, can be imputed to errors in technic which are easily averted. To present the dangers attending brachial plexus block, and a few methods of preventing them, has been the object of this paper.

SUMMARY

This article, based on a series of 3000 cases, treats of the dangers attending brachial plexus block. It completes a study of this method which was begun with a former publication. We prefer the use of 20 cc. of novocain (2 per cent) without adrenalin. The supraclivacular technic of Kulenkampff is followed, and the dangers attending the infraclavicular technics of Balog, Anglada, Santoni, and the axillary technic of Hirschel, are pointed out. A consideration of meralgias with paresthesias as observed by different authors has been given. To three cases of death following brachial plexus block gleaned from the literature, we add our own case which is of a different origin. We indicate methods of relieving accidents and of obviating them. We believe this type of anesthesia is preferable for all surgery of the upper extremity.

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FEMORAL HERNIA: A TECHNIC OF REPAIR*

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THE CLINIC

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It has been asserted that there have been more than one hundred methods described for repairing femoral hernias. This fact seems to indicate that no one method has proven to be universally satisfactory. The method of repair to be described in all probability has been used before but, if so, I have been unable to find reference to it in the literature. If this method has been published it has not been given the publicity it deserves and this is my reason for adding to the already voluminous literature covering the subject.

There are two schools of thought regarding the extent of the operation necessary for cure of femoral hernia. First, those who believe that high ligation and extirpation of the sac is all that is necessary and, second, those who believe that, in addition to this, an attempt should be made either to obliterate the femoral canal or to close off its proximal opening through which the hernia has protruded. It is my belief that the second premise is the one more nearly correct.

It is very difficult if not impossible to determine just how frequently recurrences do occur following any type of repair, first, because femoral hernia is not a common condition and any one surgeon does not have the opportunity of following a large series of cases in which he has used a given technic of repair and, second, because it is difficult to get an accurate follow-up of the cases operated upon over a long period of time.

Shelley's[1] report on the patients with hernia observed at St. Luke's Hospital, New York, from 1916 to 1935, inclusive, is the largest series I have been able to find. This included 238 femoral hernias which comprised 2.35 per cent of all hernias seen. One hundred and forty of these, after primary repair, were examined postoperatively for nine months or longer. The recurrence rate was 3.6 per cent. Thirteen cases with secondary repair showed a recurrence rate of 15.4 per cent.

Dickson[2] comments as follows: "Most writers believe that femoral hernia is a simple surgical problem with satisfactory results. A smaller group believes that the recurrence rate is high and the true results of operation hard to estimate because of inadequate follow-up. Recurrences have been stated by various authors at rates of from 4 to 30 per cent. Extremely low figures are probably incorrect because of the difficulty of following cases after operation."

Ochsner,[3] in 1906, wrote, "It is a well-known fact that it is practically impossible to keep a circular opening in any part of the body from closing

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spontaneously unless it be lined with a mucous or a serous membrane." Therefore, in the usual femoral hernia repair he advised only cleaning out the femoral canal and high ligation and excision of the sac. When one considers how rigid the anatomic structures surrounding a femoral hernia are, it is difficult to understand how such an opening can close spontaneously.

Russell[4] in 1926, discussing femoral hernia, reached these conclusions: "Closure and abolition of the sac by separation and firm torsioning from below is in my opinion by far the safest and simplest and best method of dealing with it."

More recently, in 1942, Waugh and Hausfeld[5] recommended simple excision of the sac without closing the femoral ring or canal.

Many other references could be cited favoring the simple type of repair.

On the other hand, the majority of surgeons advocate, in addition to high removal of the sac, some procedure directed at obliteration of the femoral canal or closure of its proximal opening. Such pioneer observers as Bassini, Ferguson, Halsted, Moschkowitz, Andrews and a host of others since their time have contributed various methods of bringing this about.

Suturing Poupart's ligament to the pectineus fascia or to Cooper's ligament (Shelley[1]) has been a favorite method. Filling the femoral canal with fat derived either from resected omentum or from the abdominal wall, using the resected sac, or filling the canal with catgut are methods that have been described. Bone pegs, staples, screws, transplanted fascia, and osteoplastic flaps from the pubes have had their advocates. The femoral opening has been closed by utilization of the transversalis fascia (Dickson[2] and Harkins and Swenson[6]) by turning in a flap of the external oblique aponeurosis (Andrews[7]), by suturing the conjoined tendon to the pectineus fascia or Cooper's ligament (Lotheissen[8]) and by other methods too numerous to recount at this time.

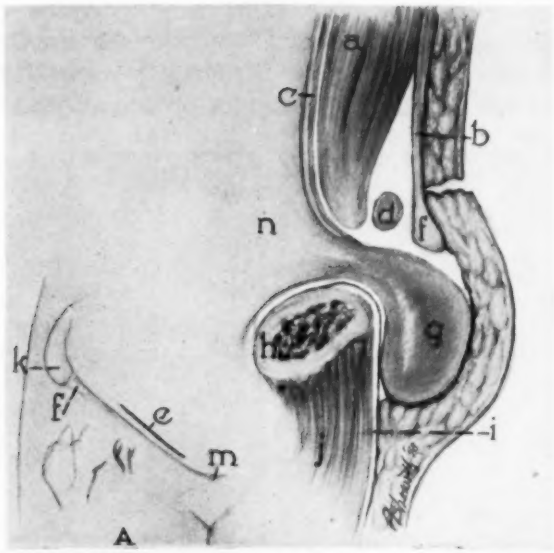
Attempts at closing the femoral canal by suturing Poupart's ligament or the conjoined tendon to the pectineus fascia or Cooper's ligament have the common weakness of suturing tissues under tension and in all probability in the majority of instances those structures in time return to their original positions, leaving the femoral canal as patent as before its attempted closure.

Transversalis fascia is a valuable adjunct to the repair of both inguinal and femoral hernias, when this structure is sufficiently developed but unfortunately it commonly is least evident when most needed. The same may be said of the aponeurosis of the external oblique and the pectineus fascia so it is well to have in mind several methods of procedure that may be applicable to the individual case.

There are two methods of approach commonly used in the repair of femoral hernias; one below Poupart's ligament and the other above, using an incision similar to that used in the repair of an inguinal hernia. The latter method of exposure, in almost everyone's experience, is far preferable to the former, though a combination of the two may be desirable, particularly when dealing with strangulated intestine.

Before taking up the details of the method of repair of femoral hernia to

FIG. 1



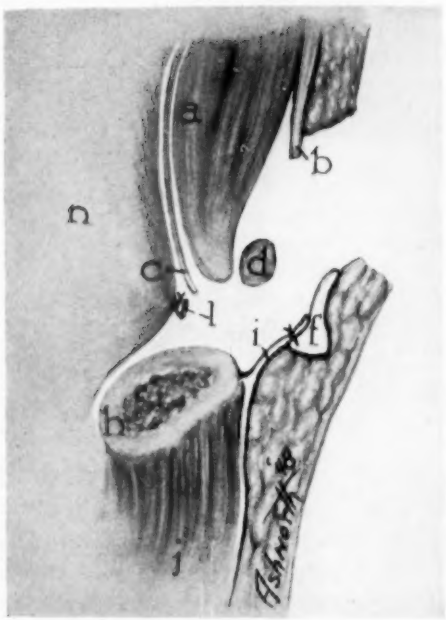


FIG. 2

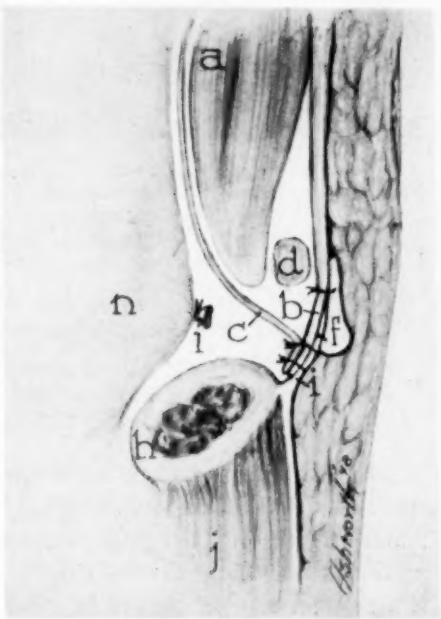


FIG. 3

FIGS. 1, 2, 3.—Drawings showing the incision, sagittal section of a femoral hernia with its anatomic relations and the technic of using these structures in its repair.

a. Internal oblique muscle
b. Aponeurosis of external oblique muscle
c. Transversalis fascia
d. Round ligament
e. Incision
f. Poupart's ligament
g. Hernia sac
h. Pubic bone
i. Pectineus fascia
j. Pectineus muscle
k. Anterior-superior spine of ilium
l. Neck of sac ligated
m. Spine of pubis
n. Peritoneum

be described, it will be advantageous to review briefly the anatomy of the parts concerned.

The femoral ring and femoral canal as such do not normally exist since the femoral vein lies in contiguity with Gimbernat's (lacunar) ligament. However, when a femoral hernia develops by the protrusion of abdominal content,

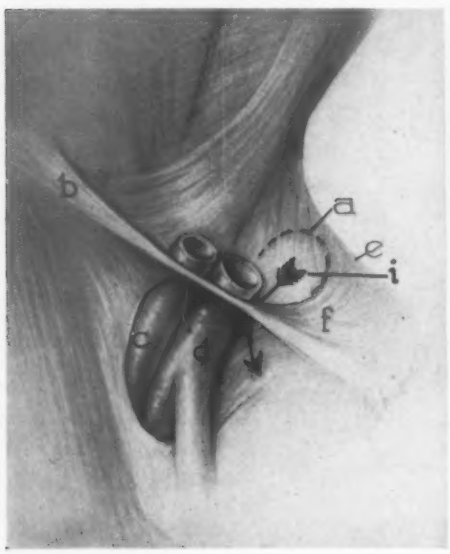


FIG. 4

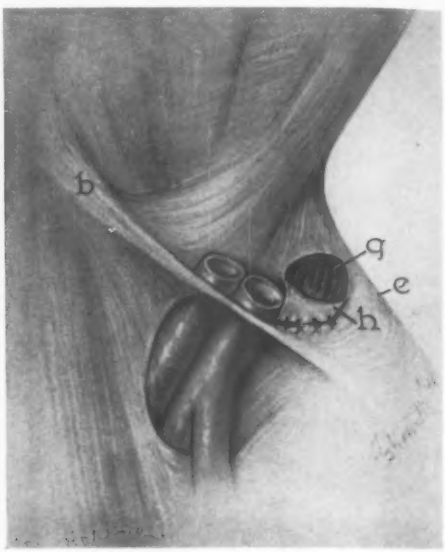


FIG. 5

FIGS. 4, 5.—Drawings of anterio-posterior view to show the method of using pectineus fascia to close the femoral canal.

a. Incision into pectineus fascia
b. Poupart's ligament
c. Femoral A
d. Femoral V
e. Cooper's ligament covering pectineal line
f. Gimbernat's ligament
g. Pectineal muscle
h. Flap of pectineal fascia
i. Femoral canal

usually small gut or omentum or both, the peritoneum is pushed through the space medial to the femoral vein and lateral to Gimbernat's (lacunar) ligament. Anteriorly, lie Poupart's ligament and transversalis fascia and posteriorly, the pectineus fascia and Cooper's ligament. Such a canal, when fully developed, is approximately ¾ inch in length and extends from the femoral ring above to the fossa ovalis below.

Poupart's ligament, as is well known, is a strong fibrous band extending from the anterior superior spine of the ilium to the pubic tubercle and is continuous with the aponeurosis of the external oblique muscle above. Medially near the attachment of the ligament to the pubic tubercle, diverging fibers are given off which pass inward and upward to the pectineal line, thus giving rise to Gimbernat's ligament.

Cooper's ligament is a strong fibrous band which is continuous with Gim-

bernat's ligament and extends laterally along the pectineal line to which it is attached. It is strengthened by and continuous with the pectineal fascia which overlies the pectineus muscle. From anatomic dissection and from observation of the attachments of these ligaments to the pelvic bones, it is evident that Cooper's ligament occupies a plane proximal to Poupart's ligament and this becomes more pronounced as the pectineal line swings laterally. This fact permits a flap of pectineus fascia to be dissected up and sutured to Poupart's ligament without tension (Figs. 4, 5).

Transversalis fascia, when well developed, is a tough fibrous structure which lies just superficial to the peritoneum and through which both direct and indirect inguinal hernias, as well as femoral, must pass. Needless to say, such hernias cannot develop so long as this fascia remains intact. It is an extremely valuable tissue for use in the repair of these hernias when well developed but it is frequently found to be an entirely inadequate structure.

The aponeurosis of the external oblique muscle is attached to the superior margin of Poupart's ligament throughout its entire length. In the majority of instances, it is a well developed, tough, fibrous structure and is easily mobilized over a wide area so that usually it can easily be brought down to Cooper's ligament without tension. When well developed, it is valuable in the repair of both inguinal and femoral hernias but it also frequently is such an attenuated structure as to be of little value.

THE OPERATION

It is necessary to make the inguinal type of exposure if the method to be described is used.

An incision is made parallel to and slightly above Poupart's ligament, two thirds of the distance from the pubic spine to the anterior-superior spine of the ilium. The aponeurosis of the external oblique muscle is incised just above Poupart's ligament. The cord, or round ligament, is retracted out of the way. The neck of the femoral sac is freed, the sac opened and contents, if any, reduced. The sac is then dissected from the femoral canal, ligated high and excised. The femoral vessels are then retracted laterally. The pectineus fascia and Cooper's ligament are cleansed of attached cellular tissue. An incision is made into the pectineus fascia to include Cooper's ligament and a flap of these structures fashioned sufficiently large to cover over the femoral canal and to permit its being sutured to the inner side of Poupart's ligament without tension (Fig. 1, 5). It is surprising how easy it is to obtain such a flap and what an adequate structure it proves to be in most instances. None of the pectineus muscle is included. A search is made for the transversalis fascia and when found it is carefully dissected out and mobilized and sutured to the inner side of the flap of the pectineus fascia and laterally to Poupart's ligament along with the aponeurosis of the external oblique muscle. The lower edge of the external oblique aponeurosis attached to Poupart's ligament is sutured to the adjacent fascia as illustrated in the accompanying diagrams. This repair adequately deals with the femoral hernia and takes care of the

potential formation of an inguinal hernia that might result from such an incision.

This type of repair has been found to be particularly advantageous when it has been found necessary to sever Poupart's ligament to reduce strangulated gut when severing Gimbernat's ligament did not give adequate enlargement to the entrance to the femoral canal. Interrupted silk or cotton sutures are used throughout the repair.

ADVANTAGES OF THE OPERATION AS DESCRIBED

As has been pointed out, it is my belief that in addition to the removal of the sac in femoral hernia an attempt should be made to either obliterate the femoral canal or close off the proximal femoral opening in order to minimize recurrences. Utilization of the transversalis fascia or the aponeurosis of the external oblique muscle serves this purpose well when these structures are well developed and when they can be sufficiently mobilized to be sutured to Cooper's ligament without tension. With sufficient mobilization of these structures this can usually be accomplished.

Pectineus fascia, on the other hand, is readily accessible with a minimum of dissection necessary for its mobilization. It is a well developed structure and it can be sutured to Poupart's ligament without tension; or, at least, such has been the case in those individuals subjected to operation and in dissections on the cadaver, with one exception, in which case the fascia was very poorly developed.

It is well in dealing with hernias to have in mind several alternate procedures, for it has been everyone's experience that certain structures for the repair of hernia may be well developed in one individual and utterly lacking or attenuated in others. In other words, fit the operation to the individual and not the individual to the operation.

This presentation has the same fallacies as most discussions on the subject. The method has not been used sufficiently often nor results observed over a sufficiently long period of time to arrive at definite conclusions. In my experience with 606 hernia repairs of all kinds, 20 were for femoral hernia. Five of these were repaired by the technic described. However, the method appears to be anatomically sound and I believe wider use will substantiate this belief.

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RESULTS OF TREATMENT OF PERFORATION OF THE ESOPHAGUS*

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FROM 1925 THROUGH 1947, 69 cases of peri-esophageal infection following perforation of the esophagus by instrumentation, foreign body, or both were encountered at the Mount Sinai Hospital, New York City. The results in the first 40 of these (to mid-1941) have already been published by Neuhof and Jemerin. However, as the number of cases in the original report has subsequently been augmented by almost 75 per cent, and as the factor of chemotherapy must now also be evaluated, an inclusive report at this time seems pertinent.

Of the 69 patients, 44 recovered and 25 died, a gross mortality of 36.2 per cent. Our interest, however, lies in the results of treatment, and gross mortality is therefore meaningless. The important thing is to determine whether, with the passage of years, results have improved. If so, can this improvement be correlated with a better understanding of the sequelae of perforation and with methods of treatment? In Table I the cases are tabulated according to years.

From the table, it is readily seen that definite progress has been made. Dividing the cases into two groups, those prior to 1936 and those since, the progress is even more apparent. For, of the 22 patients from 1925 through 1935, 17 died, a mortality of 77.3 per cent, while of the 47 patients since 1936, only eight died, a mortality of 17.0 per cent. The reason for selecting 1936 as the dividing point is not arbitrary and will be apparent later.

To what is this striking difference in mortality attributable? In part, the answer is supplied by further dividing the cases into those operated upon and those treated conservatively. Sixteen patients were not operated upon. Of these, 11, or 68.7 per cent died. Fifty-three patients had definitive operative procedures directed toward the lesion produced by the perforation as distinct from palliative procedures such as gastrostomy or tracheotomy. Fourteen, or 26.4 per cent of these died. By breaking down the material further into a chronologic listing of the operative and non-operative cases, additional information is obtained in Table II.

It is now seen why 1936 has been selected as the dividing point. At about this time, surgery began to be recognized as the treatment of choice. Prior to this date, only 50 per cent of the cases were treated surgically, while about 90 per cent have been accorded its benefits since. Thus, the improvement in results has paralleled the increased utilization of surgical therapy. However, other factors must certainly have been at play also as the results for surgical therapy itself have improved markedly. This is shown by the reduction in

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mortality for surgically treated cases from 72.7 per cent before 1936 to 14.3 per cent since that date.

The most important single factor instrumental in recovery is the recognition of esophageal perforation as a surgical lesion. The next most important is realization of the need for prompt intervention. From its inception, peri-esophageal infection following esophageal perforation is almost always diffuse and intense in nature. This is because usually the bacteria are

TABLE I.—*Cases of Esophageal Perforation. Chronologic Listing of Recoveries and Deaths.*

Year	No. of Cases	Recoveries	Deaths
1925	1	0	1
1927	2	1	1
1928	1	1	0
1929	1	0	1
1930	4	0	4
1931	6	1	5
1932	2	1	1
1933	2	0	2
1934	1	0	1
1935	2	1	1
1936	5	4	1
1938	6	5	1
1939	5	2	3
1940	3	3	0
1941	7	6	1
1942	2	1	1
1944	4	3	1
1945	4	4	0
1946	7	7	0
1947	4	4	0
	69	44	25
			Gross mortality 36.2%
Cases prior to 1936:	22	5	17
			Mortality 77.3%
Cases since 1936:	47	39	8
			Mortality 17.0%

anerobic and because they are introduced too suddenly for local defenses to be mobilized. Prompt destruction of tissue with liquefaction resulting in rapid abscess formation usually occurs and the process is not infrequently putrid or gangrenous. But a wide zone of cellular inflammation remains surrounding the abscess. If the inflammation is not checked by drainage or other means, this serves as a zone of spread for a mediastinal phlegmon or cellulitis which was almost always the common denominator in the fatal cases.

Obviously, it is not the perforation *per se* which kills but the infection which spreads in the highly cellular cervico-mediastinal planes. Thus, in the usual case, drainage must be prompt and adequate in order for it to have its greatest chance. Analysis of the case material indicates that in early years operation was performed reluctantly, almost as a last resort measure, while in recent years prompt drainage has been the rule. Prior to 1936, in only three

of the 11 surgically treated cases was operation performed less than one week after the perforation had occurred. Since 1936, 35 of the 42 treated surgically were operated upon less than a week after perforation and 20 of these were operated upon within two days.

TABLE II.—*Cases of Esophageal Perforation. Chronologic Listing of Operative and Non-Operative Recoveries and Deaths.*

Year	No. of Cases	Operated			Non-Operated		
		No.	Recovered	Died	No.	Recovered	Died
1925	1	1	0	1	0	0	0
1927	2	0	0	0	2	1	1
1928	1	0	0	0	1	1	0
1929	1	1	0	1	0	0	0
1930	4	2	0	2	4	0	2
1931	6	3	1	2	3	0	3
1932	2	2	1	1	0	0	0
1933	2	1	0	1	1	0	1
1934	1	0	0	0	1	0	1
1935	2	1	1	0	1	0	1
1936	5	4	4	0	1	0	1
1938	6	6	5	1	0	0	0
1939	5	5	2	3	0	0	0
1940	3	3	3	0	0	0	0
1941	7	7	6	1	0	0	0
1942	2	2	1	1	0	0	0
1944	4	3	3	0	1	0	1
1945	4	4	4	0	0	0	0
1946	7	4	4	0	3	3	0
1947	4	4	4	0	0	0	0
	69	53	39	14	16	5	11
		Operative Mortality 26.4%			Non-operative Mortality 68.7%		
Cases prior to 1936: 50% treated surgically; only 3 out of 11 operated upon less than 1 week after perforation.							
	22	11	3	8	11	2	9
		Operative Mortality 72.7%			Non-operative Mortality 81.8%		
Cases since 1936: 89.4% treated surgically; 35 out of 42 cases operated upon less than 1 week after perforation; 20 cases operated upon less than 2 days after perforation.							
	47	42	36	6	5	3	2
		Operative Mortality 14.3%			Non-operative Mortality 40.0%*		

* This figure does not include many non-operative failures whose lives were saved by late surgical intervention (see text).

Ideally, operation is performed immediately upon recognition of the existence of a traumatic laceration of the esophagus and certainly before infection has descended into or become full blown in the mediastinum. Such an operation can be regarded in the nature of a prophylactic measure. However, this is not always possible. Many cases arrive only a considerable time after the accident has occurred. Fortunately deterioration is not always rapid. When

the perforating agent has been a small foreign body such as a fish bone, the resulting infection may be less virulent and develop much more slowly than after a large tear by an instrument. Also, not infrequently the process has been delayed by the spontaneous establishment of partial drainage by perforation of the abscess into the esophagus or lung. But one has only to contrast the satisfactory results of early operation with the disastrous results of non-intervention or late operation to realize that perforation of the esophagus provides an urgent indication for operation whenever possible.

Operation in itself accounts for practically no mortality. Perforation usually takes place at the junction of the pharynx with the esophagus and accordingly, in most cases, cervico-mediastinal drainage effected through the neck under local anesthesia is sufficient to take care of the lesion. The technic, patterned after Marschik, is simple and well standardized. It is described in detail by Neuhof and Jemerin. The operation was performed in 41 of the 53 patients in our series that were operated upon and only in three cases was it necessary to supplement the procedure by a subsequent posterior mediastiotomy. Of the remaining 12 operative cases, primary posterior mediastinotomy was performed in three because of the location of the lesion, and thoracotomy with drainage of mediastinal and complicating lung abscess or empyema was performed in the other nine. Needless to say, appropriate supportive measures to achieve optimum general condition are an integral part of the surgery.

Analysis of the deaths in the operative cases before 1936 shows that these were due for the most part to spread of infection because of too long delayed or improperly conceived operation. The characteristic mode of spread in these fatal cases was as mediastinal phlegmon. Since 1936, the six deaths following surgery were due to secondary hemorrhage from major vessels in two cases, cardiovascular collapse in one case in which the true nature of the basic condition was not recognized until the patient was in extremis, delayed poorly conceived operation in one case, and overwhelming infection in one case not admitted until ten days after perforation.

The non-operated patients died, of course, of undrained infection. One of the two patients since 1936 was admitted in extremis with infection already spread throughout the mediastinum. In the other, the course of events following esophagoscopy was not recognized as the sequelae to an esophageal perforation and death ensued despite sulfadiazine and penicillin.

The recent availability of various potent chemotherapeutic agents again raises the question of conservative therapy. Actually, it must be recognized that an effort was made to treat conservatively many cases that came to operation, or at least to observe them for a sufficient period to determine whether they would resolve without operation. Thus the 53 operative cases include 40 in which there was a preliminary period of waiting of at least two days (either before or after they were admitted), and 25 in which the period of temporization was at least five days. The longest interval before operation was 10½ months in a case of mediastinal abscess which had perforated into the lung. Since 1939 most of these cases have had chemotherapy, the sulfona-

mides prior to 1944 and penicillin with or without the sulfonamides since 1944. The course seemed to be in no way influenced by the sulfonamides but in 1946, three patients recovered with penicillin therapy. However, one died which would otherwise have been saved and five other patients with preliminary penicillin for five, three, one, nine, and two days, respectively, deteriorated while under treatment and required surgery. One of these deteriorated so rapidly despite penicillin that in two days she was almost moribund and her recovery after operation was a surprise. Another was discharged apparently well after five days of penicillin only to return a week later with an abscess that required drainage. The three patients who recovered with penicillin alone were apparently minor perforations with only minimal peri-esophageal contamination. Thus it is seen that the failures with non-operative therapy are not given by the mortality figures alone. The mortality under conservative therapy including the antibiotics would have been much higher had not many of these cases been taken out of that category and saved by operative intervention.

Streptomycin has not had an adequate trial but our impression is that in esophageal perforation no chemotherapeutic agent is as yet a substitute for prompt drainage, particularly since we are dealing with anerobic infection in a closed space and an operative procedure in itself so little hazardous. Only potential harm can be caused by waiting, and, in general, the more prompt the operation, the greater the chance for recovery. Actually, mortality from esophageal perforation should be rare, occurring only in unpreventable situations such as a patient admitted with too widespread an infection late after perforation, fulminating infection following extensive laceration of the esophagus, and serious complicating lesions.

SUMMARY

1. The results in 69 cases of esophageal perforation covering a period from 1925 through 1947 are presented.

2. Chronological listing of the results shows a marked improvement with the passage of the years.

3. Further breakdown into operative and non-operative cases shows the improvement to be due to an increased incidence of prompt surgical intervention.

4. Penicillin and the other chemotherapeutic agents should be used only as an adjunct to prompt surgery.

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TREATMENT OF PANCREATIC CYSTS*†

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PANCREATIC CYSTS may be of the proliferative or the non-proliferative type. Though the literature is scant, most proliferative cysts reported have been malignant.[1] Extirpation of this type of cyst is the treatment of choice because it represents a precancerous lesion. Nonproliferative cysts, on the other hand, rarely if ever undergo malignant degeneration and therefore can be treated in one of several ways. Extirpation should be employed if the cyst can be easily enucleated. If the cyst is fused intimately with surrounding structures, its removal might prove hazardous and increase unnecessarily the mortality rate. In the past, marsupialization has been employed. Gussenbauer[2] in 1883 performed the first external drainage of a pancreatic cyst. Judd[3] reported 33 cases of marsupialization, many of which were followed by sinus tracts that persisted for two years. Kerr[4] had a patient who drained for 15 years. The objections to this form of therapy are autodigestion of the abdominal wall, infection of the cyst and sinus tracts, severe inanition and repeated hemorrhages. Attempts to close these fistulae by irradiation,[5] radium,[6] and sclerosing agents[7] have not proven efficacious. Marsupialization should have little place in our present surgical armamentarium.

Another therapeutic approach, which has an increasing number of proponents, is the performance of an anastomosis between the cyst and a hollow viscus. The first anastomosis was performed by Jedlicka,[8] between the cyst and stomach. Anastomoses between the cyst and the gallbladder have been reported by Waltzel[9] and Neuffer[10] with variable results. Failures have occurred probably because of contamination of the cyst by bacteria and intestinal contents.

Recently Adams[11] reported two cases of anastomosis between the jejunum and pancreatic cyst with an auxiliary jejuno-jejunostomy. Both these patients had satisfactory convalescences which he attributed to successful shunting of the jejunal contents away from the lumen of the cyst. To increase the likelihood of this diversion, a Roux-en-Y anastomosis was performed in one of our cases.

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Case 1.—A. P., a 27-year-old white male, entered the hospital on April 11, 1947, complaining of severe left lower quadrant and para-umbilical pain. Twenty-eight hours before admission he first became aware of para-umbilical pain which radiated to the left lower quadrant. Although the pain was constant, there were occasional severe colicy attacks. There was no nausea, vomiting, diarrhea or melena. The urinary tract was negative.

Examination revealed an acutely sick male lying with his thighs maintained in flexion. Temperature was 99° F., pulse 92 and respirations 20. The abdomen was scaphoid. Marked tenderness and spasm were elicited over the left rectus abdominis muscle. Hyperperistalsis was present. Rectal examination was uninformative. The white blood cell count was 15,400. The differential smear was normal. Serum amylase was 8 units (normal 8-64 units.) Intravenous pyelography revealed a displacement of the left ureter toward the midline, obliteration of the left psoas shadow and probable indentation of the lesser curvature of the stomach. The patient's symptoms became more severe and three hours after admission laparotomy was performed.

Operation: The abdomen was opened through a left rectus muscle-splitting incision. A large pancreatic cyst was found lying in the lesser omental cavity with a thumb-like projection that protruded behind the stomach through the gastrohepatic omentum. This projection was markedly hyperemic and covered with fresh fibrin. There was no evidence of fat necrosis or free fluid in the abdomen. After exposing the cyst through the gastrocolic ligament, the finger-like projection was freed from the gastrohepatic omentum and was excised. A large amount of necrotic debris and serosanguineous fluid was evacuated from the cyst which comprised the entire body and tail of the pancreas. Because the cyst was firmly adherent to the base of the transverse mesocolon, the splenic vessels and the posterior wall of the stomach, extirpation was deemed unwise. The jejunum was then divided about 10 inches from the ligament of Treitz. The aboral end was delivered through an opening in the mesocolon and was anastomosed to the dome of the cyst with a two-layer technique. The oral end of the jejunum was anastomosed to the distal loop about 6 inches from the cysto-jejunostomy in the manner of Roux.

The fluid evacuated from the cyst was found to contain pancreatic ferments and a pathological diagnosis of pseudocyst of the pancreas was made.

Clinical course. The patient made an uneventful convalescence. A gastro-intestinal series on the 12th postoperative day revealed a normal intestinal pattern. There was no filling of the cyst cavity. The patient was discharged on the 14th postoperative day. A gastro-intestinal series taken two and six months postoperatively revealed no abnormalities. He has continued to be asymptomatic.

COMMENT

Although the follow-up of this patient has been short, the advantages of an internal anastomosis over marsupialization are evident in the patient's asymptomatic convalescence. The anastomosis en-Y should prove more effective than a cysto-jejunostomy in continuity, by virtue of side-tracking the intestinal content away from the cyst.

Case 2.—R. L. D., a 54-year-old male, entered the hospital for the fifth time on April 1, 1947, complaining of nausea and vomiting of two months' duration.

He was first admitted to the hospital in December 1941, because of episodes of gastric distress, pain and flatulence. After the gastro-intestinal and gallbladder series were reported negative, he was discharged. He returned in September 1942, because of weakness, weight loss and jaundice. On October 17, 1942, a laparotomy revealed a large cyst of the head of the pancreas which was drained externally. In March, 1944, he entered the hospital for the third time because of persistent drainage from the sinus tract. Surgical excision of the cyst and sinus tract was attempted, but the surgeon was unable to remove

the cyst in toto. A new sinus developed postoperatively and continued to drain after he left the hospital. This sinus tract closed spontaneously in January, 1945. In March, 1945, he entered again and an incisional hernia was successfully repaired.

He felt well until February, 1947, when he began to have nausea and vomiting which became progressively worse. He entered another hospital where he was treated conservatively for intestinal obstruction for 20 days. He left the hospital only to return four days later with the same complaints. After a few days he was symptomatically better and was discharged. Forty-eight hours later, his symptoms recurred and he entered the hospital on April 1, 1947, with a diagnosis of intestinal obstruction.

Physical examination revealed a thin, undernourished, slightly jaundiced, chronically ill male. A mass the size of a large orange, which appeared to be firmly adherent to the deep structures, was felt in the epigastrium. The red blood cell count was 3,600,000; hemoglobin 12 Gm.; white blood cell count 10,000 with 79 per cent neutrophils. The icterus index was 28. Roentgenograms of the chest revealed a right apical opacity which had increased in size since previous roentgenograms taken in 1941. There was also an increase in the number and size of emphysematous blebs in the right lung since 1941. Sputum and gastric examinations revealed no acid fast bacilli.

Because of the persistence of jaundice and the patient's downhill course, laparotomy was performed on April 19, 1947, through a transverse upper abdominal incision dividing both recti. A large cyst, measuring 10 cm. in diameter, was found involving the head and body of the pancreas. The gallbladder was tense, appeared normal, but could not be emptied by manual compression. The cyst was exposed and opened. After draining the amber colored fluid, a line of cleavage was finally established between the cyst wall and capsule. The cyst was removed completely. The gallbladder was then easily compressible and was emptied. It was inferred that extirpation of the cyst relieved compression of the common duct. No injury to the common duct was noted. A rubber Penrose drain was left in the lesser omental cavity and the abdomen closed in layers.

The fluid from the cyst contained pancreatic ferments. The pathological diagnosis was cyst of the pancreas, either retention or pseudocyst.

Clinical course. On the first postoperative day the patient had a spontaneous pneumothorax, and at this time tubercle bacilli were found in the sputum. Pneumothorax was continued with weekly refills.

On the fourth postoperative day, biliary drainage was noted from the operative wound. Stools became clay colored. Lipiodol injected into the fistula entered the duodenum. Patient continued to have epigastric distress relieved on occasion by probing the sinus tract with a release of bile-stained material and a concomitant diminution in the icterus index. On May 12, 1947, a catheter was inserted into the sinus tract. Diodrast outlined the entire biliary tree and the catheter appeared to be in the common duct. Patient was prepared for further surgery.

The second exploration was accomplished on July 7, 1947, with the catheter still in the common duct. A side-to-side choledochoduodenostomy, as advocated by Sanders,[12] was performed. A T-tube was inserted into the common duct above the site of the anastomosis. This was brought out through a separate stab wound in the flank.

The postoperative course was uncomplicated. On the 36th postoperative day, the T-tube was removed after repeated lipiodol injections revealed the dye entering the duodenum. The original sinus wound drained less and less bile-stained material, and finally closed. The icterus index became normal. He had no complaints when last examined in October, 1948. The pneumothorax refills have been continued without incident.

COMMENT

This patient with a long history of a pancreatic cyst and draining fistulae underwent extirpation of the cyst. Unfortunately, a biliary fistula developed, which required further surgery. Since bile was not noted on the dressing

until the third postoperative day, it is probable that the lower part of the common duct damaged by the previous compression of the duct by the cyst had been eroded by the pancreatic ferments postoperatively. In retrospect, an anastomosis en-Y, as described in Case I, would have been preferable.

DISCUSSION

The incidence of carcinoma associated with proliferative pancreatic cysts is so high that the complete excision of these cysts should be performed. In the treatment of the non-proliferative type of pancreatic cyst, the decision is less clear cut. Marsupialization in general is a poor operation because of its many untoward sequelae: erosion of the skin, infection of the sinus tract, and failure of the fistula to heal. The operation of choice is either an internal anastomosis between the cyst and the jejunum or the extirpation of the cyst wall. The latter procedure should be performed whenever possible, but it is contraindicated when its employment might jeopardize the integrity of important contiguous structures. If jaundice is present preoperatively as in Case 2, excision of the cyst is unsafe because of the high incidence of injury to the common duct, either at the time of operation or by enzymatic erosion postoperatively. Carter and Slattery[13] reported injuring the common duct in two of the five cases treated by excision of the cyst. The rent in the duct was closed and a T-tube inserted into the common duct proximally. These patients had a satisfactory but protracted convalescence. An internal anastomosis between the cyst and jejunum is a safer procedure in the complicated case. The morbidity and mortality for this operation cannot be properly adduced until a much larger series is compiled. The results thus far have been so gratifying that employment of this procedure seems warranted. Infection of the cyst wall should be a less frequent complication, if the intestinal stream is shunted away from the cyst by the performance of a cysto-jejunostomy en-Y rather than by anastomosing those two structures in continuity.

SUMMARY

1. Two cases of non-proliferative cysts of the pancreas have been operated upon successfully.

2. One was treated by excision of the wall, the other by an anastomosis between the cyst and the jejunum en-Y (Roux).

3. Non-proliferative cysts involving the head of the pancreas should not be extirpated because of the high incidence of injury to common bile and pancreatic ducts.

4. This is the first report of the successful application of the Roux type of anastomosis to the treatment of pancreatic cysts.

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BOOK REVIEW

ATLAS OF PLASTIC SURGERY, BY M. I. BERSON, M.D., NEW YORK CITY, GRUNE & STRATTON, INC.—1948

In his "Atlas of Plastic Surgery" Berson presents graphically standard methods and technics in reparative surgery. The normal anatomy and common types of deformity are depicted in line drawings and photographs. Necessary instruments and consecutive steps for repair of these deformities are likewise illustrated by photographs, drawings, and an accompanying explanatory text.

In the eight chapters into which the Atlas is subdivided, that dealing with the nose is by far the most complete and most valuable. It is recommended to any who are uncertain of indications for the several technics of corrective rhinoplasty. The presentation is simple and direct and shows an appreciation by the author of the methods, problems and pitfalls in this procedure. The chapters on the ear, eyelids and lips are likewise well done and most of the corrective technics for minor deformities of these structures are graphically presented. The operation of "face-lifting," and some of the technics of mammaplasty and lipectomy are presented in some detail. The Mirault operation for single harelip and the Federspiel operation for double harelip are shown. The methods presented for cleft palate repair are taken from the writings of Dorrance and Bransfield. The securing and use of fascia, bone, and cartilage transplants for deformities of the face are illustrated.

The very important matter of skin transplantation, both as pedicle flaps and as free grafts, is disappointingly inadequate. The author does present the Z-plastic procedure, which lends itself to illustration. Various types of skin suture and repair are shown, including closure by the somewhat time-worn impractical geometrical patterns that have been passed down from generation to generation of plastic surgeons.

For those surgeons desiring a better understanding of cosmetic plastic surgery of the face and particularly of the nose, lips, and ears, this Atlas will prove of real value. It will not be helpful, however, to those interested in general plastic surgery of massive and complicated repairs of the face and jaws and of reparative surgery of the extremities.

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PANCREATIC PSEUDOCYSTS: REPORT OF A CASE TREATED BY CYSTOGASTROSTOMY*†

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PSEUDOCYSTS OF THE PANCREAS are not true neoplasms but are collections of pancreatic secretions in the lesser peritoneal sac. They have no epithelial lining, their walls being formed by the peritoneum of the lesser sac. These collections usually present between the stomach and transverse colon, beneath the gastrocolic omentum, although they may present between the stomach and liver or between the leaves of the transverse mesocolon. Most pseudocysts follow an attack of acute pancreatitis or pancreatic necrosis, although cases have been reported following upper abdominal trauma. The interval between the original episode of pancreatitis and the clinical manifestations of the cyst is usually several weeks.

The diagnosis of pancreatic pseudocyst is usually made by roentgenologic studies after the finding of a mass in the upper abdomen developing a few weeks after an attack of acute pancreatitis. A most significant finding is distortion of the gastric or duodenal outlines by an extrinsic mass seen after the ingestion of a barium meal. Cysts in the body of the pancreas will cause indentation of the greater curvature of the stomach and downward displacement of the transverse colon and duodeno-jejunal junction. Cysts in the region of the head of the pancreas may cause widening of the duodenal loop. Intravenous urograms may show impaired function of the left kidney and incomplete filling of the left renal pelvis. These findings may be due to the pressure of the cyst on the renal vessels.[1]

Pseudocysts usually contain turbid fluid which may be hemorrhagic. Shreds of necrotic pancreatic tissue may be present. Bacterial contamination is common and is frequently due to colon organisms. High amylase readings from examined fluid prove its pancreatic origin.[2]

Excision of pancreatic pseudocysts has proved difficult, if not impossible, and has been attended by a high mortality. As a result the accepted treatment has been evacuation of the contents and marsupialization of the sac. This procedure is followed by prolonged drainage, skin excoriation, loss of fluid and electrolytes and the occasional necessity of secondary operations to close the persistent pancreatic fistula which develops in some cases. In attempting to overcome these undesirable sequelae of marsupialization various methods of internal drainage have been devised.

Internal drainage by anastomosis of the cyst to some portion of the gastro-intestinal tract has been done successfully several times. The procedures in which good results have been reported are cystojejunostomy and cystogas-

* Read before the Philadelphia Academy of Surgery on October 6th, 1947.
† Submitted for publication April, 1948.

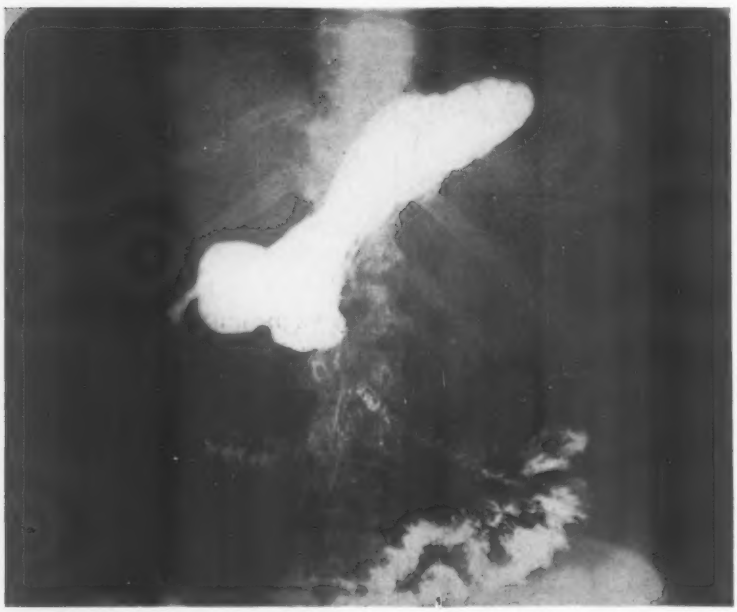

FIG. 1.—X-ray of stomach in prone position showing marked distortion and displacement of greater curvature. Taken January 28, 1947.

FIG. 2.—X-ray of colon taken on January 28, 1947 showing downward displacement of splenic flexure.

trostomy. In 1943 Chesterman[3] reported a successful case of cystojejunostomy. Adams and Nishijima[4] in 1946 reported two cystojejunostomies done at the Lahey Clinic with good results. The first recorded successful cystogastrostomy was done by Jedlicka in 1923.[5] In 1931 Jurasz[6] reported two cases done by incising the anterior wall of the stomach, making a cautery opening through the posterior wall of the stomach into the cyst and closing the anterior gastrotomy. Harries,[7] in 1934 reported a successful cystogastrostomy. Brocq and Aboulker[8] in 1940, reviewed the literature and found 17 cases in which internal drainage of pancreatic pseudocysts had been done, including cystogas-

FIG. 3.—Taken on March 3, 1947. Arrow points to barium in remaining cyst cavity.

trostomies, cystoduodenostomies, cystocholecystostomies and cystojejunostomies. These included the cases of Jedlicka, Jurasz and Harries. Mahadevan[9] in 1943 reported a case in which the anastomosis was done between the posterior gastric wall and the cyst, after doing an anterior gastrotomy. dos Santos[10] in 1944 reported a similar case. The case report which follows is one in which two separate pseudocysts developed in a female patient, one of which was treated by marsupialization, the other by cystogastrostomy.

CASE REPORT

M. H., white female, age 55, entered Jefferson Hospital, Philadelphia, on January 25, 1947, complaining of pain in the left flank, nausea and vomiting. She gave a history of an attack of vague abdominal pain beginning on December 15, 1946, followed the next

day by chills, fever, nausea, vomiting and severe epigastric pain radiating to the left shoulder. These symptoms continued with diminishing intensity for 4 days. She did not have medical attention during this time. On January 15, 1947 she began to have nausea and vomiting again. Since January 22nd she had excruciating abdominal pain, pain in the left loin and flank, chills and fever. She had no urinary symptoms or change in bowel habit.

Physical examination showed a pale, apprehensive, middle-aged female in obvious discomfort. There was marked left-sided abdominal rigidity and tenderness. It was difficult to be sure because of the rigidity but it was felt that there was a mass in the left upper abdomen. No jaundice was present. The white blood cell count on the day of admission was 10,400. Urine obtained by catheterization was loaded with red blood cells. The temperature at this time was 101.4. An intravenous urogram done on January 26,

FIG. 4.—Taken on April 3, 1947. Arrow points to fleck of barium which represents final cyst remnant.

1947 showed incomplete filling of the left renal pelvis and ureter. Cystoscopy was done on January 27, 1947. Clear urine was obtained from the left kidney. A retrograde pyelogram showed a left bifid renal pelvis but was otherwise normal. The roentgenologist reported that there was no evidence of intrinsic renal disease but that a perinephric abscess might be present. The patient was seen on January 27, 1947 by Dr. D. M. Davis whose opinon was that there was no renal or perirenal disease to account for her symptoms. On January 28, 1947 a barium meal and barium enema were given. Roentgenograms showed a mass in the left upper abdominal quadrant displacing the stomach forward and to the right (Fig. 1) and the splenic flexure downward (Fig. 2). A chest roentgenogram showed a small amount of fluid in the left pleural cavity. The tentative diagnosis of pseudocyst of the pancreas was strongly confirmed by the roentgenologic findings. A serum amylase done on January 28, 1947 was reported as 800 units. The patient was prepared and operation was carried out on January 29, 1947. The abdomen was opened through a left upper transverse incision and a large pancreatic pseudocyst was found presenting through the gastrocolic omentum. Many small areas of fat necrosis

were seen in the omentum. The cyst was aspirated and about 2 liters of straw-colored fluid containing particles of necrotic tissue were removed. The interior of the cyst was explored digitally and no loculations found. The cyst was attached to the mid portion of the pancreas. The cyst was marsupialized and the incision closed. An amylase reading from the aspirated fluid was 64,000 units. Following operation there was a large amount of drainage which was severely irritating to the surrounding skin. Difficulty was encountered postoperatively in maintaining normal fluid and electrolyte balance and in spite of intensive care the carbon dioxide combining power and plasma chlorides tended to remain low. On February 8, 1947 an ovoid mass was felt in the right upper abdominal quadrant. Clinical jaundice was noted on February 17, 1947. On February 18, 1947 the patient was operated upon again. A right upper rectus incision was made and a large pancreatic pseudocyst was found in the region of the head of the pancreas. This was making extrinsic pressure on the duodenum and the retroduodenal portion of the common bile duct. The gallbladder contained many calculi. The cyst presented through the gastrocolic omentum and was densely adherent to the posterior wall of the stomach. The common bile duct was markedly dilated. The cyst was aspirated and contained a thick brown fluid which upon culture showed growth of colon bacilli. The old transverse incision was then opened and digital exploration of the cyst and the old pancreatic fistula carried out simultaneously. No communication could be demonstrated between the two. Cholecystectomy was then done and the common duct was opened. Catheters and dilators passed freely into the duodenum and it was felt that the common duct obstruction had been due entirely to extrinsic pressure. A T-tube was placed in the common duct. A two layer anastomosis was then done between the cyst and the stomach, the anastomosis being placed on the greater curvature close to the pylorus. The incision was closed and the patient returned to her room in good condition. The jaundice had subsided completely the following day. In the next 2 weeks the patient improved greatly. Biliary drainage gradually decreased. Drainage from the old pancreatic fistula stopped almost entirely. On March 3, 1947 a cholangiogram showed immediate entrance of the dye into the duodenum and normal filling of the hepatic radicals (Fig. 3). A barium meal was then given which showed normal gastric emptying and a persistent shadow below the pylorus suggesting barium in the remnant of the cyst (Fig. 3). The patient was sent home on March 11, 1947 with the T-tube clamped. She was readmitted for study on April 1, 1947. Her general condition was excellent and she had no symptoms. There was no external biliary or pancreatic drainage. Her serum amylase at this time was 80 units. A barium enema on April 1, 1947 showed no displacement of the colon. A cholangiogram showed no obstruction to the entrance of the dye into the duodenum. Roentgenograms taken after a barium meal on April 3, 1947 showed a fleck of barium below the pylorus which was interpreted as being the last small remnant of the cyst (Fig. 4). Gastric analysis on April 5, 1947 showed normal values for free and total acid and an amylase of less than 80 units. Fasting blood sugar was 93 mg/100 cc. The T-tube was removed on April 5, 1947 and the patient was discharged. She has gained weight and has been symptom free up to the present time. Her incisions are well healed and there is no external biliary or pancreatic drainage.

SUMMARY

This case is reported because of the rarity of the development of two pancreatic pseudocysts in one patient and to demonstrate the successful use of a method of treatment of pancreatic pseudocysts which has been used very seldom in the past.

We believe that the method of anastomosis used is practical and can be applied in any case in which the cyst wall is firm enough to permit careful suturing. The major appeal of this method is the avoidance of prolonged

drainage, skin excoriation, fluid and electrolyte loss and secondary operations for the closure of fistulae which may follow upon marsupialization.

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ORAL STREPTOMYCIN IN SURGERY OF THE LARGE BOWEL:

The Production of Secondary Hypoprothrombinemia*

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STREPTOMYCIN, administered orally, is being employed with increasing frequency in several clinics in the preparation of the large bowel for surgery.[10, 12, 18] The superiority of this antibiotic over the sulfonamides, sulfathalidine and sulfasuxidine, previously employed in the reduction of the bacterial flora of the large intestine was established by Ravdin and his co-workers.[10]

It has been suggested that in the course of diminishing the bacterial population of the colon, sulfasuxidine and sulfathalidine interfere with the synthesis of an anti-hemorrhagic factor, probably vitamin K. Sanders and Halperin[13] have reported postoperative hemorrhage in two patients prepared with oral sulfasuxidine and demonstrated the concomitant occurrence of a hypoprothrombinemia in both cases. The hypoprothrombinemia in each instance was readily remedied by synthetic vitamin K preparations administered intravenously. A similar occurrence was noted on the surgical service of Montefiore Hospital in a patient prepared for an intraperitoneal colostomy closure with sulfasuxidine. On the third postoperative day this patient had a massive hemorrhage into the bowel which coincided with a markedly increased prothrombin time. Pollard,[8] however, while treating four cases of chronic ulcerative colitis with 12 Gm. of sulfasuxidine daily for three weeks, failed to find any significant evidence of disturbance in the prothrombin time. Black and his group[2] administered sulfaguanidine to rats and demonstrated a significant increase in the prothrombin time as determined on a 12.5 per cent dilution of the rats' plasma.

The purpose of the present communication is to demonstrate the appearance of hypoprothrombinemia, as manifested by an increase in prothrombin time, when streptomycin is administered orally to reduce the colonic bacterial count.

METHODS

The prothrombin time was determined by the method described by Shapiro[14, 15] which was based on the technics of Link and his co-workers.[2] Determinations were done on whole and 12.5 per cent diluted plasma specimens as suggested by these investigators.

Bacterial counts were obtained from stool specimens employing nutrient glucose beef infusion agar plates with serial dilutions of wet triturated stool;

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the results were read as viable bacteria per gram of wet stool. The preparation of the stool for culture followed the method proposed by Poth.[9] The bacterial counts from normal stools as obtained in the present study were strikingly lower than those reported in the papers reviewed.[9, 10] Nevertheless, the decrease in the bacterial counts that was noted after ingestion of the drug served the purpose of the study.

Four normal subjects were employed. The prothrombin times and the bacterial count in the stool were determined for each subject on two different days prior to the institution of oral streptomycin. Each subject was thereupon given one Gm. of streptomycin in a glass of tap water three times daily, and this regimen was continued for 14 days. The prothrombin time and the stool bacterial count were determined 24 hours after the inception of streptomycin by mouth and then at 48- to 72-hour intervals thereafter for 14 days.

No attempt was made to modify the diet of these subjects either as to type of food or its quantity. Two subjects received the regular hospital ward diet; the others continued their usual extra-hospital diets. It was suggested that each dose of the drug be taken by the subject one hour prior to each meal; this routine was adhered to in varying degrees by each of the four people studied.

DISCUSSION

The oral administration of streptomycin produced a diminution in stool bacterial count varying in the four cases from a 100 per cent reduction in one instance (Subject I) wherein culture of the stool revealed no growth to an 80 per cent reduction in Subject III who proved to be the most refractory. It is evident that the antibiotic varies in its effectiveness in different individuals as it does in its bacteriostatic action on different pathogens. In all subjects the maximum reduction in stool bacterial count was achieved within 48 hours of the institution of the streptomycin. Concomitant with the decreased bacterial counts in the stool, there appeared a significant elevation in the prothrombin times of both whole and diluted plasma specimens. The peak of the hypoprothrombinemia as measured by the maximum whole plasma prothrombin times was reached within one to four days; the maximum dilute plasma prothrombin times were reached within two to seven days. The increment in prothrombin time in dilute plasma was greater than that noted in the whole plasma specimens. This observation appears to be in agreement with the finding of Black *et al*[2] who suggested the use of dilute plasma prothrombin time determinations as a more sensitive means of detecting reductions in prothrombin levels in the blood.

In all subjects, there appeared to be an escape from the effect of the streptomycin; the bacterial counts began rising more or less progressively in every case within four to seven days after the drug was started. In three subjects, E. coli alone appeared in increasing numbers; in Subject III streptococci appeared first in great numbers and were then overgrown by streptomycin resistant coli bacilli. Roughly paralleling this increasing bacterial count was the rising level of prothrombin as evidenced by decreasing prothrombin times so that by the end of two weeks both dilute and whole plasma prothrom-

bin times approached pre-streptomycin values. This escape phenomenon is probably explained by the development of drug fastness. Several investigators have noted that bacteria rapidly acquire resistance to doses of streptomycin which were initially bacteriocidal.[7, 12] In the present study the development of bacterial resistance despite seemingly lethal concentrations of the antibiotic in the bowel lumen was possibly due to the presence of relatively inaccessible foci, such as the mucosal crypts of the colon. The appearance of drug fastness was irreversible; in all instances the bacterial counts remained high in the second week of treatment with streptomycin. The persistence of resistant bacterial strains in these individuals is of clinical importance. Resistant strains were found in patients treated for chronic urinary tract infections several months after the cessation of streptomycin therapy and in one instance Murray and his group demonstrated resistant bacteria some 11 months later.[7]

In addition to the development of drug fastness, the problem of uniform admixture of the drug with the bacterial substrate should be considered. Small quantities of the antibiotic at frequent intervals are probably more effective than more massive doses at longer intervals. Similarly, decreasing the fecal bulk by feeding a low residue diet should enhance the effectiveness of the streptomycin by allowing more immediate and prolonged contact between the intestinal bacteria and the drug.

It appears that the plasma prothrombin times are roughly inverse functions of the stool bacterial count. Almquist[1] has shown that numerous organisms such as *B. proteus, E. coli, streptococcus lacticus,* and *B. aerogenes* synthesize vitamin K when cultured on a vitamin K-free nutrient medium. The vitamin K manufactured by the coliform bacteria in the animal intestine, and readily absorbed in the presence of bile, probably accounts for the infrequency with which simple nutritional deficiency of vitamin K occurs in man.[6] It is possible that the hemorrhagic disease of the newborn which has been directly attributed to a demonstrable hypoprothrombinemia, occurs prior to the advent of a colonic flora in the newborn infant's sterile large intestine and, therefore, before the establishment of the endogenous source of vitamin K in the body. In the four subjects studied, once the remaining bacteria became drug fast and reproduced their kind, the synthesis of vitamin K approached a normal level of activity and the secondary hypoprothrombinemia disappeared.

In this study, no systemic toxic effects, other than the hypoprothrombinemia, were noted; this would seem to bear out the work of previous investigators[7, 10, 12, 16] who have demonstrated minimal absorption, if any at all, of streptomycin from the intestinal tract, with recovery of 90 per cent of the drug dose (given orally) in the stool.

Two subjects spontaneously remarked on the appearance of a sweet taste in the mouth after one day on streptomycin; the abolition of oral fetor was noted objectively in these two subjects. All four of the subjects noted a diminution in the gross bulk of the stool with a decided decrease in the fecal odor.

Oral streptomycin has been employed clinically in eight cases on the surgical service in the past six months in this hospital. All eight patients had

TABLE I: *Results*

SUBJECT I

Date	Stool Bacterial Count (Colonies per Gm. of Wet Stool)	Prothrombin Time (Seconds) Whole Plasma	12.5 % Plasma
2 days pre-streptomycin	53,000 *E. coli*	17.0	34.0
1 day pre-streptomycin	49,000	17.6	38.0
1 day post-streptomycin	no growth	23.8	51.0
2 days	100	22.5	58.0
4 days	200	21.6	75.4
7 days	2,200	21.4	74.5
9 days	6,300	20.6	66.0
11 days	2,000	20.4	62.8
14 days	16,400	18.0	38.5

SUBJECT II

Date	Stool Bacterial Count (Colonies per Gm. of Wet Stool)	Prothrombin Time (Seconds) Whole Plasma	12.5 % Plasma
2 days per-streptomycin	11,000 *E. coli*	18.7	49.6
1 day pre-streptomycin	29,000	19.0	51.0
1 day post-streptomycin	200	22.3	61.3
2 days	200	22.1	56.3
4 days	1,000	22.6	60.4
7 days	2,000	22.4	72.2
9 days	4,000	17.9	60.5
11 days	8,600	18.2	49.8
14 days	3,400	18.0	51.3

SUBJECT III

Date	Stool Bacterial Count (Colonies per Gm. of Wet Stool)	Frothrombin Time (Seconds) Whole Plasma	12.5 % Plasma
2 days pre-streptomycin	32,000 *E. coli* predom.	19.8	50.8
1 day pre-streptomycin	38,000	18.6	50.4
1 day post-streptomycin	6,000	20.3	50.6
2 days	1,000	23.0	64.6
4 days	10,800 Streptococci predom.	19.9	54.0
7 days	57,000 Streptococci predom.	18.6	52.8
9 days	1,000 *E. coli* predom.	17.9	50.1
11 days	4,700	18.2	52.8
14 days	22,000	18.8	51.2

SUBJECT IV

Date	Stool Bacterial Count (Colonies per Gm. of Wet Stool)	Prothrombin Time (Seconds) Whole Plasma	12.5 % Plasma
2 days pre-streptomycin	53,000 *E. coli*	16.4	32.6
1 day pre-streptomycin	69,000	16.1	36.0
1 day post-streptomycin	300	20.8	48.6
2 days	100	22.0	51.1
4 days	3,000	21.4	49.8
7 days	10,000	18.7	40.7
9 days	21,400	17.2	36.0
11 days	9,600	19.0	40.4
14 days	18,400	17.0	38.6

proven or suspected lesions in the colon and received the drug in the course of preoperative preparation. In these operative cases, streptomycin has been administered in 0.5 Gm. doses every four hours for two days immediately preoperatively in conjunction with parenteral synthetic vitamin K. The adjuvant preparatory measures of low residue diet and mechanical cleansing by enemata have been continued as well. Where peritoneal contamination has been anticipated streptomycin has been given parenterally for two days preoperatively and two days postoperatively. The vitamin K preparations have been continued postoperatively for one week. All eight operative cases have had uneventful postoperative courses with maintenance of prothrombin time at pre-streptomycin levels. There have been no hemorrhagic complications.

SUMMARY

Reductions in stool bacterial count ranging from 80 to 100 per cent were achieved in four normal subjects by the oral administration of streptomycin. The maximum reduction in the bacterial counts in all subjects was noted within 48 hours of the institution of the streptomycin. It must be pointed out that the initial bacterial counts obtained in this study were very much lower than those reported by other authors employing the same technic.

Simultaneous with the decrease in the bacterial counts was the appearance of a hypoprothrombinemia as manifested by significant increases in prothrombin times determined in both whole and 12.5 per cent diluted plasma.

CONCLUSIONS

Where streptomycin is employed preoperatively in the surgery of the large bowel, the operative procedure should not be delayed for more than 48-72 hours after the drug is started.

It is suggested that vitamin K preparations be given before and after operation to prevent the appearance of the hypoprothrombinemia noted when streptomycin is given orally.

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ERRATUM

In the September 1948 issue of the ANNALS OF SURGERY, in the paper "The Response to Vagotomy in Idiopathic Ulcerative Colitis and Regional Enteritis" by Dr. Clarence Dennis, we failed to place a credit line with the color plate which appeared facing page 486 of this issue. This notice is as follows:

"Publication of these colored photographs has been made possible through the generosity of Mr. H. W. Rutzen, to whom the authors wish to express their thanks."

ERRATUM

In the article by Dr. Gordon Murray in the October 1948 issue of the ANNALS OF SURGERY, Figure 7 on page 850, through a typographic error, was printed upside down. The legend as printed should thus be interpreted with the illustration in reverse position to that printed. The same applies to Figure 8 on page 851 where the illustration is also upside down. The legend must also be re-interpreted in this case.

CONTROL OF THE COMMON ILIAC ARTERY DURING SACRO-ILIAC DISARTICULATION (HEMIPELVECTOMY)*

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PREVENTION OF HEMORRHAGE is of particular importance during the wide dissection necessary in sacro-iliac disarticulation. This has been emphasized by Girard,[1] Gordon-Taylor,[2, 3] Judin[4] and Pack,[5] and various methods of vascular control have been described. Ligation of the external iliac artery, necessary in the removal of the extremity, does not control the parietal branches of the hypogastric artery encountered in the division of the pubic and sacro-iliac synchondroses and the structures deep in the pelvis. Momberg's procedure of temporary occlusion of the abdominal aorta is dangerous and unnecessary. Preliminary ligation of the common iliac artery achieves the necessary vascular control and makes the operative procedure relatively bloodless. However, the occurrence of necrosis of the posterior flap of the amputation following ligation of the common iliac artery is frequent. Judin[4] states that Brzosovski reports five patients who developed posterior flap necrosis after common iliac artery ligation and that Krassintzev's patient developed a necrotic flap after a similar ligation. Pack[5] reports this complication and states that such ligation should not be done routinely but "may be rarely indicated in poor risk patients where excessive bleeding, shock and prolonged operation are to be avoided at all costs."

Temporary control of the common iliac artery during the operative procedure will achieve the benefits of ligation without any of its undesirable effects. Any suitable type of direct arterial tourniquette may be employed. We have used the form of arterial control described by Babcock.[6] In the early stage of the operation, after division of the insertion of the rectus abdominis muscle and the attachments of the inguinal ligament, the peritoneum is retracted medially and the external iliac, hypogastric and common iliac arteries identified and a controlling tape placed around the common iliac artery. The ends of the tape are passed through the lumen of a short segment of soft rubber tubing and are drawn tight, compressing the artery against the tube. The tape is held taut by clamping a hemostat across the tube. (Fig. 1.) Occlusion of the common iliac artery is maintained throughout the operative procedure. After the hindquarter is removed, the controlling tape is released, thus preserving the continuity of the common iliac artery and its blood supply through the hypogastric artery and its branches.

Sacro-iliac disarticulation performed on three patients, with primary malignant tumors involving the hemipelvis, is herein reported. The standard technic outlined by Pack[5] with the addition of common iliac artery control

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was followed in each patient. It was observed that bleeding was far less than in previous experiences with the operation where the common iliac artery was not occluded. The absence of hemorrhage resulted in a shorter, less troublesome procedure in which shock did not occur at any stage.

CASE REPORTS

Case 1.—J. W., white, male, age 20, was admitted to the Veterans Hospital, Portland, Oregon on January 17, 1947, complaining of a painful mass in the left gluteal region, first noted in November, 1946. An unsuccessful attempt at local excision had

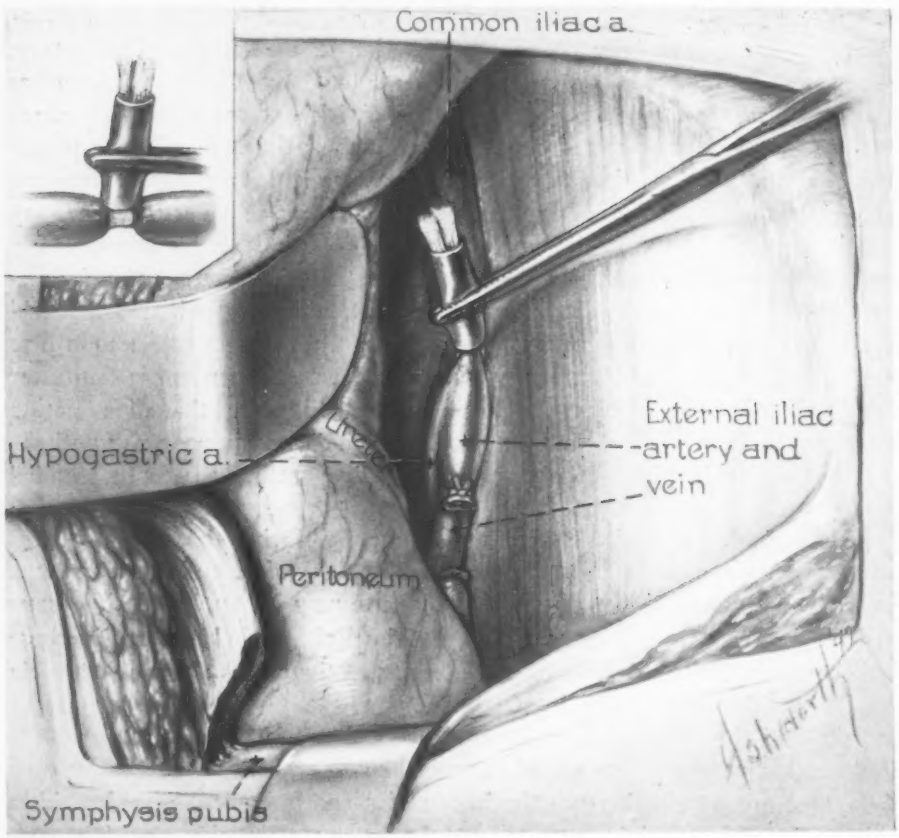

FIG. 1.—Drawing at operation illustrating method used for temporary control of the common iliac artery during sacro-iliac disarticulation.

been made at another hospital in December, 1946, after biopsy had shown the tumor to be an osteochondrosarcoma. Admission examination revealed a healed operative scar in the left buttock overlying a large, hard, tender mass. The left thigh was flexed and the patient could not bear weight on the extremity. Radiographic examination of the pelvis by Dr. Hyman revealed a dense bone condensation along the iliac margin of the left sacro-iliac joint, arising along the iliac crest. Near this placque of bone sclerosis was a group of small, circular, translucent areas of bone destruction. The appearance was

that of a chondrosarcoma. Roentgenograms of the chest, long bones and skull were negative.

Operation, March 10, 1947—Under endotracheal, nitrous-oxide, oxygen, pentothal anesthesia, an incision was made beginning at the symphysis pubis, extending across the left iliac fossa to the anterior superior spine of the ilium, continuing downward over the greater trochanter and across the gluteal fold, then upward to the point of origin. The attachment of the left rectus abdominis muscle was cut and the inguinal ligament divided at its attachments to the pubis and the anterior iliac spine. The inferior epigastric vessels were ligated and divided and the spermatic cord retracted. The iliac fossa and the retroperitoneal pelvic space were exposed by retracting the detached abdominal wall and peritoneum medially. A large, hard, nodular tumor mass filled the iliac fossa, overlying and involving the sacro-iliac joint and extending into the pubis. It had spread posteriorly through the gluteal muscles into the superficial fascia and subcutaneous tissue. The common iliac, external iliac and hypogastric arteries were exposed. A controlling tape was placed around the common iliac artery which was occluded throughout the remainder of the operation. The external iliac artery was transfixed, doubly ligated and divided. The symphysis pubis was identified and divided with a chisel. It was observed that there was only slight bleeding during this procedure.

FIG. 2.—Case 1.—Appearance of posterior skin flap, four weeks after operation.

The patient was turned on his right side and the posterior dissection was carried through the muscles attached to the iliac crest and the gluteal attachments to the posterior surface of the sacrum. Returning to the anterior dissection the psoas muscle and femoral nerve were divided and the left sacro-iliac joint exposed. The external iliac vein was now transfixed, doubly ligated and divided. The sacrum was divided medial to the sacroiliac joint. The gluteal and obturator vessels and nerves, the sciatic nerve and the piriformis and levator ani muscles were sectioned. The sacrotuberous, sacrospinous and posterior sacro-iliac ligaments were divided and the extremity removed. The controlling tape on the common iliac artery was removed. A normal pulsation in this artery, the hypogastric, and the stump of the external iliac artery was observed. The skin flaps were approximated, using through and through interrupted sutures of silk. During the operation the systolic blood pressure was never below 110.

Pathology. The ilium was the site of a large tumor mass involving a portion of the adjacent pubic bone and the sacro-iliac joint. The tumor was cartilaginous in consistency with central areas of cystic degeneration. Microscopic study revealed a typical osteochondrosarcoma.

The postoperative course was uneventful. Constant gastric suction through a Levine tube was maintained for four days. A Foley catheter was kept in the bladder until the third postoperative day. There was no abdominal distention or urinary retention following

removal of the tubes. The wound healed without infection and the posterior flap showed no necrosis. (Fig. 2.) Follow-up examination in February, 1948, one year after operation, revealed the patient to be alive but there were metastatic nodules in both lung fields.

COMMENT

Because of the widespread nature of the tumor, sacro-iliac disarticulation was purely palliative in this case, but made it possible for a bedridden patient to be ambulatory for approximately one year. Control of the common iliac artery resulted in a minimum blood loss during the division of the pubic and sacro-iliac synchondroses and the deep pelvic dissection.

Case 2.—R. T., white, male, age 38, was first seen at the Veterans Hospital, Portland, Oregon on June 24, 1947, complaining of a painful mass in the left buttock of two years' duration. During the six months prior to admission, the mass had grown rapidly. Examination revealed a hard, immovable, firm ovoid mass in the left gluteal region. Radiographic studies of the pelvis showed areas of bone sclerosis and bone destruction within the body of the left ilium, extending to the sacro-iliac joint, characteristic of a chondrosarcoma. A biopsy of the tumor, taken on June 26, 1947, showed it to be an osteochondrosarcoma.

FIG. 3. — Case 2. — Postoperative photograph. Note primary healing.

Operation, July 21, 1947. A left sacro-iliac disarticulation was performed in the usual manner with temporary occlusion of the common iliac artery during the procedure.

Convalescence was uneventful save for a left lower lobe atelectasis which cleared after tracheal suction. There was a minimal amount of serious drainage from the wound, which was firmly healed in 21 days with no evidence of necrosis of the skin flap. (Fig. 3.)

Pathology. The specimen consisted of the left leg with the ilium, sacro-iliac joint, pubis and ischium. A firm, rounded tumor mass, 9 x 7 x 5 cm. arose from the external surface of the ilium, extending to the sacro-iliac joint. It appeared to be encapsulated, but greyish-brown strands extended from the tumor into the overlying muscles. A cross section of the tumor showed the presence of cystic spaces, partly filled with necrotic tissue. The area of the ilium from which the tumor rose showed patches of bone destruction and bone eburnation. On histologic examination, the tumor gave the characteristic picture of osteochondrosarcoma.

Follow-up examination, February, 1948, revealed a well-healed, painless stump. Roentgenograms of the remaining bones of the pelvis and the lungs showed no evidence of metastasis.

COMMENT

Sacro-iliac disarticulation may result in prolonged arrest of the malignancy in this patient; because the tumor was well circumscribed, it had not involved the sacro-iliac joint and the dissection was wide of the involved ilium. Temporary occlusion of the common iliac artery provided excellent vascular control and the healing of the wound was by primary intention.

Case 3.—L. G., white, male, age 32, was admitted to the Veterans Hospital, Portland, Oregon on October 31, 1947, because of persistent pain in his left knee and thigh of four months' duration. One month before admission, the left thigh became swollen and a biopsy, taken by a private physician and studied at this hospital, revealed a periosteal type of sclerosing osteogenic sarcoma. Admission examination revealed a large tumor mass extending from the lower third of the thigh upward for a distance of 20 cm. Roentgenograms of the left femur showed broadening of the soft tissues around its lower half, within which there was a large bone tumor, characterized by extensive bony proliferation. There were many sclerotic and demineralized areas of bone within the shaft itself extending up to the greater trochanter. Radiographs of the lungs showed no metastasis.

Operation, November 11, 1947. A left, sacro-iliac disarticulation was performed and common iliac artery occlusion was maintained throughout the procedure.

Convalescence was uneventful save for considerable sero-sanguinous drainage from the wound. There was no necrosis of the flaps. (Fig. 4.)

Follow-up in February, 1948, showed a well-healed stump with no evidence of local recurrence or metastasis.

Pathology. The specimen consisted of an entire lower extremity which had been disarticulated at the pubic and sacro-iliac synchondroses. A large, irregular tumor, 20 x 15 x 10 cm. arose from the lower third of the femur and extended upward within and without the cortex to the greater trochanter. The tumor was composed of hard, cartilaginous grey-white material in which areas of hemorrhage and necrosis were present. It had invaded surrounding muscles from the knee to the greater trochanter. The microscopic diagnosis was osteogenic sarcoma.

FIG. 4. — Case 3. — Postoperative photograph. Note viability of posterior skin flap.

COMMENT

Sacro-iliac disarticulation in this patient made possible the wide removal of a large osteogenic sarcoma of the femur which had extended to the greater trochanter both within and without the bone.

SUMMARY AND CONCLUSIONS

The value of temporary control of the common iliac artery is described in the report of three cases of sacro-iliac disarticulation. This addition to the standard technic of the operation makes possible a safer procedure with minimal blood loss. The danger of necrosis of the posterior skin flap incident to common iliac artery ligation is avoided.

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CORRECTION

DR. I. S. RAVDIN *has requested that the* ANNALS OF SURGERY *publish the following correction:*

"My attention has been called to a statement which I made in "The Changing Scene in American Surgery," ANNALS OF SURGERY, **127**: 666–675, 1948 to the effect that candidates for examination in plastic surgery must now be certified by the American Board of Surgery. This is an error."

I. S. RAVDIN.

ADAMANTINOMA OF THE MAXILLA METASTATIC TO THE LUNG*

CASE REPORT

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The adamantinoma (ameloblastoma) is a rare tumor most commonly found in the jaw. It grows slowly, usually over a period of many years. This epithelial tumor arises either from the remnant of the enamel organ or from cell rests scattered along the length of the tooth. It is closely related to the dentigerous cyst and might well be called an enamel cell tumor. Enlargement of the bone, with rarefaction and thinning of the cortex, occurs as a direct result of the growth of the tumor, which may be primarily either solid or cystic, although degeneration of the solid type to form pseudo-cysts is common.[1]

The literature reveals that 80 to 85 per cent of adamantinomas arise in the mandible and about 15 per cent in the maxilla. Isolated case reports indicate other primary sites such as the tibia, the lip, the cheek, or even the pituitary body. The tumor has been generally considered to be benign, but metastases to other organs have been reported.

Chont[2] briefly reviewed the literature in October 1943 and reported one additional case of distant metastasis from an adamantinoma of the mandible. There have been reported 18 cases in which distant metastases to various sites have occurred; in nine of these the metastatic lesion involved the pulmonary parenchyma (Table I). Schweitzer and Barnfield,[3] reporting also in October 1943, describe a case wherein metastasis to the lung occurred from an adamantinoma of the mandible. These authors mention the fact that in only one previous instance, that case reported by Vorzimer and Perla, was there histologic proof of the ameloblastomatous nature of the metastatic lung tumor. It is reasonable to assume that the clinical impressions of the nature of the metastatic deposits in the pulmonary parenchyma were adequate to substantiate these diagnoses in spite of the lack of histologic proof. Indeed, in that case reported by Chont, the nature of the metastatic lesion was not verified histologically, but the clinical history leaves little doubt as to its true nature. However, the fact remains that of the nine reported cases of adamantinomas metastasizing to the lung, only two have been verified histologically. We are reporting a third.

CASE REPORT

Mrs. M. C., a 56-year-old, white housewife, entered the University of California Hospital December 5, 1947. She was referred to us by her physician, Dr. Sidney Shipman. Sixteen months previously (September 1946) she had noted the onset of malaise and easy fatigability. Chest roentgenograms taken at that time demonstrated a soft shadow in the right lung field which was interpreted as bronchitis. The slight irritative

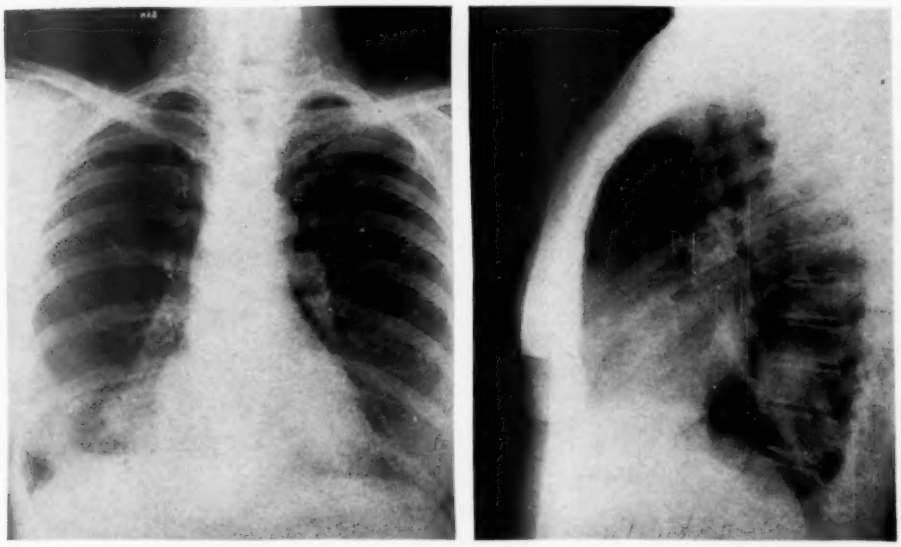
* Submitted for publication, February 1948.

TABLE I.—*Summary of Reported Metastases from Adamantinoma*

Author of Case Report	*Site of Primary Tumor*	*Duration of Primary Tumor*	*Sex*	*Age*	*Race*	*Sites of Metastases*	*Remarks*
1. EVE	Left Mandible	13 Weeks	F	60	W	Lumbar lymph nodes	
2. EWING	Unknown	Unknown		Unknown		Cervical lymph nodes	
3. EWING	Unknown	Unknown		Unknown		Lung	
4. GENTSCH	Left Maxilla	Unknown	M	44	W	Lymph nodes, sphenoid sinus, and malar bone	
5. HAVENS	Unknown	Unknown		Unknown		Unknown	
6. HAVENS	Right Mandible	Unknown	F	49	W	Right submaxillary region	
7. HEATH	Both Mandibles	28 Years	M	67	W	Local recurrence, metastases to deltoid fossa and pelvis	
8. HORSLEY	Right Mandible	20 Years	M	39	W	Local recurrence, metastases to cervical lymph nodes and lung	
9. NEW	Mandible	Unknown		Unknown		Submaxillary gland	No recurrence after excision of primary tumor & involved submaxillary gland
10. SIMMONS	Left Mandible	15 Years	F	37	W	Recurred locally, 4 times; metastasized to cervical lymph nodes.	
11. SIMMONS	Right Mandible	15 Years	M	37	W	Local recurrence, metastases to cervical, submaxillary lymph nodes & lungs	
12. SPRING	Right Mandible	Unknown	M	5	W	Recurred 7 times, metastases to cranial bones, subcutaneous tissue of forehead	Metastases disappeared with X-radiation
13. VORZIMER AND PERLA	Right Maxilla	31 Years	M	38	W	Recurred 5 times, invasion of antrum & nasal septum & metastases to lung	Metastases to lung histologically proved.
14. WEISSENFELS	Left Mandible	14 Years	M	26	W	Recurred, metastases to cervical lymph nodes, skin, submaxillary lymph nodes, & lung	
15. CHONT	Left Mandible	8 Years	F	55	W	Local recurrence, metastases to lungs	Punch biopsy of lung. Tumor not adequate for diagnosis. No autopsy.
16. OKINOUYE	Mandible	Unknown	M	15	W	Metastases to lungs	Original lesion: ameloblastoma. Metastatic lesion: resembled carcinoma.
17. PHELPS	Maxilla	Unknown	M	42	W	Metastases to liver, ribs; tumor found within pulmonary vessels	
18. SCHWEITZER AND BARNFIELD	Mandible	Unknown	F	23	C	Recurred locally despite 24 operations; involved the maxilla & calvarium & metastasized to lungs	Histologically proved.
19. GRIMES AND STEPHENS	Left Maxilla	3 Months (Approximate)	F	46	W	No local recurrence, metastasis to lung	Histologically proved.

cough, without production of sputum or blood, persisted without change until her entry into this hospital. On only one occasion, while walking hurriedly, did the patient note right anterior chest pain. She received symptomatic medical therapy. There was no weight loss. Repeat roentgenograms in July 1947 and in September 1947 each showed an increase in the size of the lesion over that of previous roentgen examinations. Because of ankylosis of the temporomandibular joints, two attempts to perform bronchoscopy were unsuccessful.

In November 1937, ten years previously, an adamantinoma of the left maxilla was removed surgically. The operative procedure was preceded by an extraction of the left upper third molar tooth resulting in a chronically draining area which was finally removed with the subjacent tumor mass. She received postoperative radium therapy which resulted eventually in a partial ankylosis of the temporomandibular joints, some of which she had overcome.



A B

FIG. 1.—Roentgenograms taken in November 1947 illustrating the increase in size of the right lower lobe lesion over that of July 1947. The circumscribed nature of the mass is maintained. It is more apparent on the lateral view than in the previous films.

Major system inventory was non-contributory in respect to the pulmonary lesion. Progressive bilateral eighth nerve deafness had been present since the age of 18, requiring the constant use of a mechanical hearing aid.

Physical examination revealed a well nourished white female, not acutely ill, with normal temperature, pulse and respiration. The blood pressure was 126/72. The left upper alveolar ridge and the adjacent maxilla, as well as the floor of the left antrum of Highmore, had been surgically removed. The posterior aspect of the left half of the hard palate also had been removed, allowing one to view a rather large cavity in the roof of the mouth extending posteriorly along the base of the skull for a distance of approximately 6 to 7 cm. A well-fitting dental prosthesis effectively closed off these openings. The mucosa surrounding the previously involved area, although exhibiting considerable scarring, was completely smooth and without evidence of recurrence of the original tumor. There were no palpable lymph glands or other masses in the cervical region.

Expansion of the chest was equal bilaterally. There was an area of decreased breath

sounds, overlaid with an exactly similar area of dullness, in the right lower lung field posteriorly. There was no evidence of an intrapleural collection of fluid.

Examination of the abdomen, pelvis and rectum revealed only normal findings throughout. Urinalysis showed normal constituents, and the hematologic study showed no anemia.

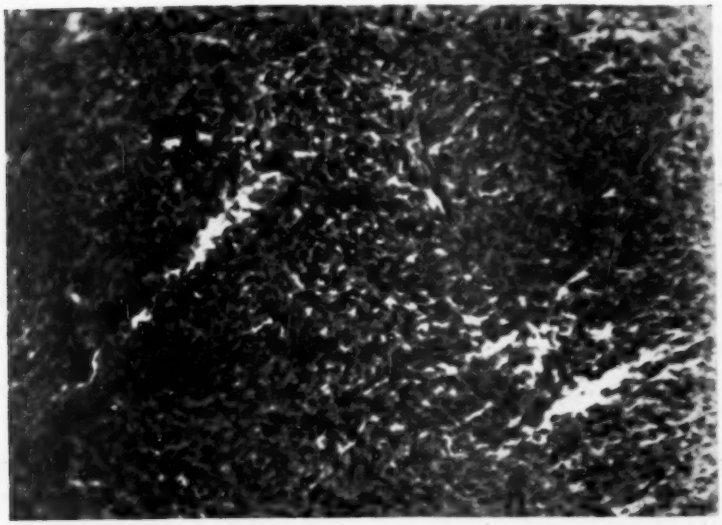
Roentgenograms of the chest (November 1947) showed a circumscribed density of the lower lobe of the right lung occupying its posterior basal division. Comparison with the films taken three and five months previously (July and September 1947) showed that the lesion had again definitely increased in size. (See Figure 1.)

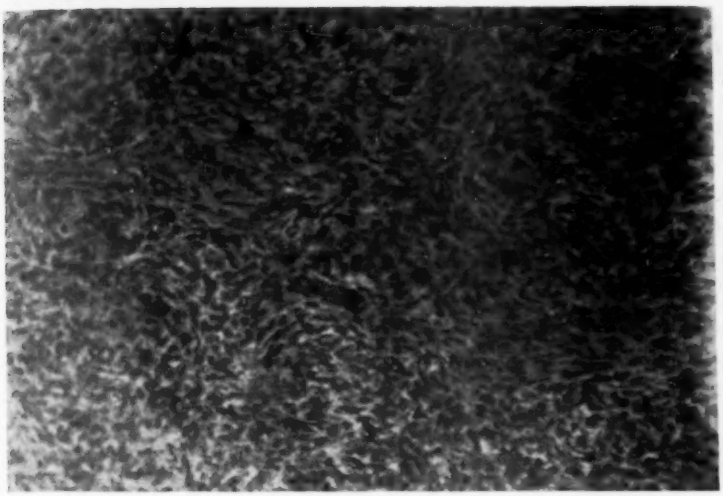
FIG. 2.—Photograph demonstrating the cut section of the tumor. The lower lobe bronchus (top left), though immediately adjacent to the mass in one area, is not involved by it. The surrounding pulmonary parenchyma is only slightly compressed and is not otherwise abnormal.

It was our opinion that we were dealing with (1) a slowly growing, low-grade primary malignancy of the right lower lobe, (2) a metastatic tumor from a primary lesion in one of the major systems not demonstrable by the usual clinical methods, or (3) a metastatic tumor from the original focus in the left maxilla.

Thoracotomy was carried out December 6, 1947. The hard, circumscribed rounded mass lay deep in the parenchyma of the right lower lobe and was entirely confined within it. The hilar and mediastinal lymph nodes in the area of dissection were soft,

A





B

FIG. 3(A)—Photomicrograph of the original lesion of the left maxilla. The cells are closely packed together and exhibit ovoid or spindle-shaped nuclei but only scanty cytoplasm. In some areas there are whorls, in others suggestive glandular arrangements. FIG. 3(B)—Photomicrograph of the metastatic tumor of the lung. Note the dense cellular pattern corresponding to that of the original lesion. The similarity between the two lesions is marked. There is apparently no correlation between the histologic pattern of the adamantinoma and its ability to metastasize.

pigmented, and were not grossly involved by abnormal tissue. There were no notable adhesions. The nature and characteristics of the tumor made one think of a secondary metastatic deposit. A right lower lobectomy was therefore accomplished. The postoperative course was entirely uneventful and the patient was discharged to her home on the 11th postoperative day.

The excised lobe of the lung contained in its center a firm, round, well circumscribed mass 3.5 cm. in diameter which was homogeneous and pinkish-white on cut section. The surrounding lung tissue showed little evidence of compression, and in general was quite well aerated. No communication between the mass and a bronchus could be demonstrated. (See Fig. 2.)

Microscopically, the tumor was composed of closely packed masses of large cells with ovoid or spindle-shaped nuclei and scanty eosinophilic cytoplasm. The cells resembled basal cells, and in some areas were palisaded at the margin. There was invasion of the surrounding connective tissue by the malignant cells. In some areas the cells were arranged in whorl-like patterns suggestive of rosettes. There was a tendency toward glandular pattern in a few areas, and occasionally the cells resembled columnar epithelial cells. In most areas, however, the masses of tumor cells were separated from the pulmonary alveoli by a heavy layer of connective tissue. Microscopic diagnosis: Adamantinocarcinoma. (See Figs. 3A and 3B.)

DISCUSSION

There is general agreement that local recurrence of an adamantinoma frequently occurs, though distant metastases are rare. Of the approximately 500 adamantinomas reported in the literature, those 18 which metastasized regionally or distally represent 3.6 per cent, while those metastasizing to the lung represent 1.8 per cent of the total.

Ewing[4] considered all adamantinomas to be malignant because of their tendency to recur and to invade bony structures in the immediate neighborhood. Many writers on the subject agree that local recurrence is the rule rather than the exception. However, some explain the high rate of local recurrence by the fact that the tumor may infiltrate the bone more than is apparent to the naked eye so that incomplete removal may be followed not by actual recurrence due to inherent malignancy, but rather by persistence of the tumor.

Distant metastases, however, can be explained only by the malignant nature of the primary ameloblastoma. The three possible routes of transfer to the lung are (1) by inhalation through the tracheo-bronchial tree during the course of an operative procedure, (2) via the lymphatic chains, and (3) by blood transport. It is reasonable to assume that in the case being reported the tumor tissue reached the lung by way of the blood stream, since involvement of lymph nodes in the neck or mediastinum was not observed, nor was there an intimate association of the tumor with a bronchus.

The fact that metastatic adamantinomas are likewise slow growing is well borne out in this case. It is probable that the metastasis occurred before or during the period of treatment of the original tumor in 1937, since no evidence of tumor tissue has subsequently been noted in the region of the left maxilla.

SUMMARY AND CONCLUSION

A case history is presented in which an adamantinoma of the left maxilla metastasized to the right lung. Histologic verification of the nature of the

primary and secondary lesions is presented. This is the third reported case of metastatic adamantinoma of the lung substantiated by microscopic examination. Although the adamantinoma is characterized mostly by local recurrence, in a small percentage of cases there are distant metastases.

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ANNOUNCEMENT OF VAN METER PRIZE AWARD

The American Goiter Association again offers the Van Meter Prize Award of Three Hundred Dollars and two honorable mentions for the best essays submitted concerning original work on problems related to the thyroid gland. The Award will be made at the annual meeting of the Association which will be held in Madison, Wisconsin, May 26th, 27th and 28th, 1949, providing essays of sufficient merit are presented in competition.

The competing essays may cover either clinical or research investigations; should not exceed 3000 words in length; must be presented in English; and a typewritten double spaced copy sent to the Corresponding Secretary, Dr. T. C. Davison, 207 Doctors Building, Atlanta 3, Georgia, not later than March 15th, 1949. The committee, who will review the manuscripts, is composed of men well qualified to judge the merits of the competing essays.

A place will be reserved on the program of the annual meeting for presentation of the Prize Award Essay by the author if it is possible for him to attend. The essay will be published in the annual Proceedings of the Association. This will not prevent its further publication, however, in any Journal selected by the author.

SEVERE CRUSHING INJURY TO THE CHEST*

REPORT OF A CASE HAVING EXTENSIVE BILATERAL RIB FRACTURES SUCCESSFULLY TREATED BY PERICOSTAL SKELETAL TRACTION

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THE AUTOMOBILE HAS MADE crushed chest a comparatively common and not infrequently fatal accident. Cardio-respiratory embarrassment due to the development of hemothorax and pneumothorax may be an outstanding feature in these injuries. More commonly, however, symptoms of this type are delayed 48 to 72 hours and are produced by a clinical triad: intrabronchial hemorrhage, ineffectual cough, and a type of anoxia which has been described in another communication.[1] An exceptionally severe injury producing multiple bilateral rib fractures may be followed immediately by grave cardio-respiratory embarrassment due to violent paradoxical movement of the chest wall. The latter disability may be of such magnitude as to require prompt correction as a life saving measure. The case report which follows is presented as an example of this type. The injuries sustained by this patient include:

(1) Bilateral anterior fractures of ribs 1-7 inclusive.
(2) Fractured skull.
(3) Compound fracture of the right radius and ulna.
(4) Compound fracture of the right tibia and fibula.
(5) Kidney injury productive of hematuria for 48 hours.

Complications developing during the course of treatment include:

(1) Left hemiplegia.
(2) Hemopneumothorax.
(3) Massive subcutaneous emphysema.
(4) Acute dilatation of the stomach.
(5) Fecal impaction.

REPORT OF CASE

M. G., a white male shoemaker, age 51, was injured in an automobile accident on November 21, 1946, when he drove his car into a tree. His companion riding in the front seat was killed instantly and the patient was thrown against the steering wheel. On admission to the hospital one hour later he was unconscious, cyanotic and pulseless. Compound fractures of the right forearm and the right leg were present. Extensive bilateral rib fractures (Fig. 1) could be diagnosed by inspection, and paradoxical movement of the sternum and anterior chest wall was of extraordinary violence. The sternum appeared to be depressed almost to the bodies of the dorsal vertebrae with each inspiratory effect.

The fractured arm and leg were splinted; oxygen was administered by B. L. B. mask; an infusion of plasma was started; and the patient was taken to the operating room. Bronchoscopy without anesthesia was done, and a small amount of bloody secretion

* Submitted for publication, April 1948.

was aspirated. This was not followed by any improvement in his condition. The anterior chest wall was then quickly prepared for operation, and proceeding again without anesthesia, an incision about 3 inches long was made over the left third rib near its cartilaginous junction where the paradoxical movement was most violent. When the rib was exposed it was grasped with a large towel clip just medial to the fracture site. Strong traction on this instrument reduced by approximately 50 per cent the paradoxical movement which now became most severe at a lower level on the same side. A second incision was then made over the left fifth rib and it was grasped medial to the fracture

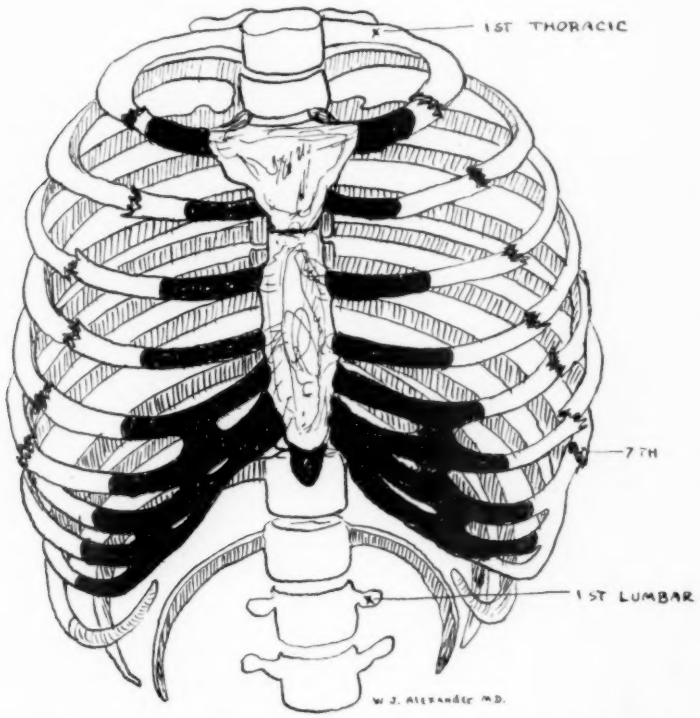

FIG. 1.—An accurate drawing which illustrates the fractures.

site with another towel clip. Strong traction on both instruments now resulted in reduction of the paradoxical movement by approximately 75 per cent and appeared to lock the fragments of the fractured ribs on the right side. The patient's breathing immediately became less labored and his color improved.

The wounds were closed and the patient was then transferred to a fracture bed. Traction was maintained by suspension to an overhead frame using rubber tubing and wire, as illustrated in Figure 2. The foot of the bed was elevated about 18 inches. Maximum stability of the chest was obtained by changing slightly the direction and the amount of traction. A transfusion of citrated blood was started and oxygen was administered by both nasal catheter and B. L. B. mask. During the first 24 hours 1,000 cc. of plasma and 1,000 cc. of blood were given. He required catheterization and the urine was bloody.

Shock was rapidly corrected but the patient remained unconscious. Accumulation of bloody tracheobronchial secretions productive of increased respiratory embarrassment followed within the first six hours after his injury. This was relieved by tracheal

aspiration using a 16 F. catheter. Clearing the tracheobronchial tree by this method was required thereafter at two to six hour intervals for the entire period of 24 days during which the traction was maintained.

The morning following injury the patient was semiconscious; his color was improved but some subcutaneous emphysema had developed. The urine was bloody. Frequent tracheal aspirations were done during the day and a large amount of bloody mucus was

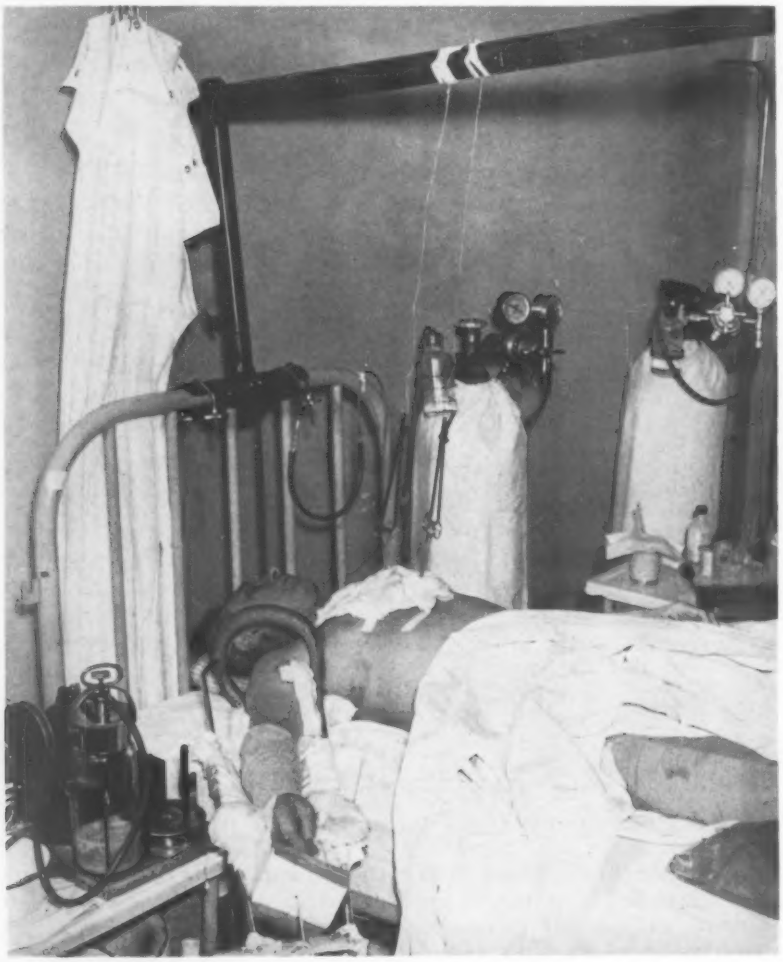

FIG. 2.—Photograph illustrating the application of traction, which was estimated at ten pounds. Note the suction machine which was kept constantly at the bedside.

aspirated. In spite of this measure and the administration of oxygen by both B. L. B. mask and nasal catheter, dyspnea and cyanosis increased alarmingly. A bedside roentgenogram demonstrated an enormous dilatation of the stomach. A Levine tube was passed which resulted in a prolonged audible escape of air. At the same time the upper abdominal wall flattened; this was followed by remarkable improvement.

The subcutaneous emphysema steadily increased in amount and on the third day of his admission it had progressed to a degree sufficient to give the skin of the anterior chest

wall a tenseness similar to that of an under-inflated football. The patient was now again alarmingly dyspneic and cyanotic. About 3,000 cc. of this air were removed, using a large needle and a pneumothorax apparatus. Deflation of the emphysema was followed by immediate relief of symptoms, but the air continued to accumulate, doubtless due to the presence of a bronchial fistula. An incision in the axillary area 2 inches long was then made into this air-containing space. This permitted continuous escape of air for the next 24 hours, after which no further leakage or accumulation occurred.

Administration of penicillin, 25,000 units every three hours, was begun at the onset. After the third day, 50,000 units were instilled twice daily through the intratracheal catheter after completion of aspiration. The bronchial secretions which were first bloody and later purulent, decreased in amount and became mucoid in character after the first week.

Seven days after admission a definite left hemiplegia had developed. He was fairly alert, nevertheless, and at this time he was able to take a liquid diet. A neurosurgeon was asked to examine the patient and on December 11th (19th day of admission) an exploratory burr hole over the right motor area was made* and an intracerebral hematoma was found and aspirated. This operation was performed without moving the patient from his bed. The procedure was well tolerated and no immediate change in his condition followed. Within three days, however, considerable improvement in his sensorium occurred and steady improvement followed thereafter. A low grade septic type of fever, present from the first, continued for 17 days although the compounded fractures remained free of infection.

Increased stability of the chest permitted some decrease in the amount of traction after the second week and oxygen therapy was discontinued. On numerous occasions during the first and second weeks, the traction was momentarily released. When this was done severe paradoxical movement and labored respiration followed immediately, and the patient became dusky and panicky after about 30 seconds. After three weeks the chest wall was only slightly mobile. After 24 days the traction was entirely released and the following day the towel clips were removed. The operative wounds healed without infection. At this time the patient developed nausea and severe abdominal cramps. Rectal examination showed the presence of a fecal impaction. This was removed with prompt relief of his abdominal symptoms.

Subsequently, open reductions of the fractured forearm (January 16, 1947) and right leg (February 4, 1947) were done.** Beginning eight weeks after injury some movement of the left leg was possible and thereafter improvement of this extremity was rapid. Five months after injury slight movement of the left arm and hand was possible and considerable additional improvement in this extremity has since occurred. Bony union of the fractured right arm and right leg followed operation. At the time of the patient's discharge from the hospital (June 29, 1947) he had a hemiplegic gait, but with a little assistance was able to walk with a cane.

DISCUSSION

In general, comparatively few patients with crushed chest injuries will require skeletal traction. The indication for this treatment arises in an occasional, exceptionally severe, injury of this type in which paradoxical movement of the chest wall is the outstanding clinical feature. The latter disability is prejudicial to normal intrathoracic pressures and thereby impairs both the function of ventilation of the lungs and the filling of the heart. The cardio-respiratory embarrassment which follows is directly proportional to the violence of the paradoxical movement.

* Operation by Dr. J. Worden Kane.

** Operation by Dr. John T. Kane.

Before skeletal traction is applied it should be carefully determined in every instance that the paradoxical movement is not contributed to, or actually precipitated, by other complications. The writer has observed several instances of severe crushed chest injuries in which paradoxical movement of the chest wall was a delayed complication. All of these patients had either extensive single unilateral rib fractures, or unilateral double fractures of several adjacent ribs. In every instance the chest wall was stable at the onset. Paradoxical movement occurred several hours or several days after injury when dyspnea developed due to hemothorax, pneumothorax, or bronchial obstruction. In these patients, immediate and dramatic relief from both the dyspnea and paradoxical movement followed the aspiration of blood or deflation of air from the pleural space or the clearing of blood and secretions from the tracheobronchial tree.

From the foregoing it is seen that skeletal traction is indicated in a small group of chest injuries in which the fractures of the thoracic cage alone are sufficient to produce paradoxical chest wall movement productive of grave cardio-respiratory embarrassment. For practical purposes it appears that few, if any, patients will require such treatment unless severe paradoxical movement is present immediately after the injury. It seems likely that traction will rarely be required in instances of single unilateral rib fractures. Such treatment may occasionally be necessary in those having unilateral double fractures of several adjacent ribs. As illustrated in this report, extensive bilateral rib fractures near the sternum which allow mobilization of a large portion of the chest wall may present an urgent indication for skeletal traction. Simplicity, and absolute reliability, are features which recommend the pericostal towel clip method with elastic overhead traction described in this case report.

A survey of the literature shows that the use of pericostal skeletal traction in crushed chest injuries has been proposed by Eloesser[2] and Wise.[3] In a personal communication, Eloesser reports the use of this type of traction in several cases but no published case report has been found in which this treatment was applied. In the comparatively few cases in which other forms of skeletal traction or fixation are reported, a delay of several hours or several days is noted between the time of injury and the time that such treatment was instituted. None of these had extensive bilateral anterior fractures near the sternum which permitted early violent paradoxical movement. This indicates that the chest injury sustained by the patient reported here may be one of unprecedented severity to have been followed by recovery.

Treatment of a fractured sternum with traction has been reported by Joslow[4] who used a coat hanger. McKim[5] reported a similar case which he immobilized with Kirschner wires. The Drinker Respirator was utilized successfully in a severe crushed chest injury with bilateral fractured ribs by Hazen.[6] Jones and Richardson[7] used skeletal traction applied to the sternum in a child having eight unilateral fractured ribs. The traction was applied with a "bullet forceps such as is used to grasp the cervix in gynecologic cases." The

traction was maintained with "weights and pulleys attached to a rainbow frame." Traction was removed after 48 hours without change in the child's condition. Their patient recovered.

A crushed chest injury which allows paradoxical movement of sufficient severity to require skeletal traction will invariably be accompanied by other complications. In all injuries of this type, intrabronchial hemorrhage associated with ineffectual cough productive of anoxia, atelectasis, and pneumonia should be anticipated. In the case reported here, effectual cough was impossible during the period that traction was maintained. This necessitated the frequent use of tracheal aspiration by the method of Cameron Haight.[8] This invaluable procedure yielded 2-4 ounces of mucopurulent bronchial secretions daily. After instruction, this method of treatment was effectively applied by the patient's nurses. Tracheal aspiration, together with elevation of the foot of the bed, and liberal use of penicillin, is believed to have made possible the avoidance of any pulmonary complication. The alarming symptoms which followed monetary release of the traction appear to demonstrate conclusively that he could not have been successfully treated without this form of stabilization.

SUMMARY

A crushed chest injury is reported in which violent paradoxical movement of the chest wall and severe cardio-respiratory embarrassment were the outstanding features. Treatment by a combination of skeletal traction, tracheal aspiration, and the administration of penicillin intramuscularly, and intratracheally, was followed by recovery.

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HERNIATION OF THE HEART*

THROUGH A PERICARDIAL INCISION

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THE FOLLOWING CASE is of interest because of the infrequency of the complication, the seriousness of the symptoms subsequent to the cardiac herniation and the dramatic recovery following its reduction.

The sequence of events is as follows: Mr. P., a rather frail, 53-year-old male, in fair general condition, was operated upon by us for a carcinoma of the left lung. The operation was uncomplicated except that there was a dense, broad adhesion which firmly bound the lower part of the upper lobe of the left lung to the parietal pericardium at the level of the conus arteriosis of the heart. Because of the density of the adhesion and its proximity to the tumor in the lung, we thought that the carcinoma might have infiltrated into it and for this reason decided to remove the pericardium with the affixed lung rather than cut through the adhesion. This resulted in a defect of the anterior pericardium about 2½ inches in diameter through which the conus and the base of the pulmonary artery could easily be seen.

Before closing the chest wall at the end of the operation, the advisability of suturing the pericardium was discussed, but we decided against it because closing this large defect would have constricted the outflow tract of the heart. I must admit that the possibility of a herniation at this site never occurred to us.

The pneumonectomy had been performed through a posteriolateral incision with the patient lying on his right side. At the end of the operation, the patient's condition was good, he was pink, his pulse was 90, his blood pressure was 110/90. Careful aspiration of the trachea and bronchus had been carried out during operation both by the anesthetist and once by us just before closing the bronchial stump, so that his respirations were free and easy.

After the dressings had been applied he was rolled over on his back, lifted on to the cart and placed gently on his operated (left) side. A moment after this was done, the entire picture changed! The patient went into collapse. His radial pulse became imperceptible, his blood pressure could not be obtained, he was pale and started to perspire. Thinking the condition was due to a cardiac shift, he was immediately put flat on his back and oxygen was administered through the tracheal tube, which, fortunately, had not yet been removed. The picture did not change. The pulse still remained imperceptible and the condition was still one of profound shock. A bottle of blood, which was available, was hooked to the

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indwelling intravenous needle in the ankle and its contents forced into the vein. Even after the entire bottle had been injected, the patient's condition did not improve. It was noted that there was no difficulty in getting air into and out of his right lung, nor did hurried examination show any evidence of unusual mediastinal shift. Thinking that the condition was most probably due to a massive internal hemorrhage, and realizing that the patient would die if nothing were done, he was put back on the operating table, pushed back into the operating room, dressings removed and draped ready for reopening the chest wall. The diagnosis of hemorrhage was strengthened by the aspiration of a few centimeters of sanguinoserous fluid from the catheter drainage tube. (Note—We usually do not drain the chest cavity following pneumonectomy, but did in this case because there had been innumerable adhesions between the visceral and parietal pleura over the entire posterior lung field and we felt that there might be a great deal of pleural reaction with a large effusion.)

It must have been at least 25 minutes from the sudden onset of shock to the start of the second operation.

When the chest cavity was re-opened through the former incision, the picture that presented was startling. The apex of the heart almost protruded through the incision. The heart appeared to be standing on end and pointing directly upward, that is, perpendicular to its usual position. It was entirely devoid of pericardial covering; it was dilated; its veins were greatly engorged and it was beating very feebly but at an extremely rapid rate. The diagnosis was obvious, namely, the heart had herniated through the pericardial defect. The rim of the pericardial opening was easily located and slit and the heart slid back into its normal position. After a few more twitches, it started to regain its normal form of contraction, and within less than half a minute was beating strongly. The cardiac dilatation and the venous engorgement receded quickly. The anesthetist was able to pick up the pulse at the temple within a few seconds and the first blood pressure reading after the cardiac replacement was 90/50 and soon 110/70. Inspection now of the left pleural cavity revealed nothing abnormal, a small amount of serosanguineous fluid, no signs of severe hemorrhage and the great vessels well tied off and the stump of the bronchus well buried in the mediastinum. It seemed to us that the entire episode was due to the angulation and the strangulation of the outflow zone of the heart incidental to the herniation. In order to guard against a recurrence, the opening in the pericardium was enlarged by removing practically the entire anterior pericardium.

By this time, the general condition of the patient was excellent. The wound was resutured; the dressings reapplied. The patient was gently turned on his back and no change in condition noted. He was returned to his room and kept on his back, with the backrest slightly elevated for the rest of the day. Needless to say, he was carefully watched!

His convalescence from here on was uneventful. He seemed to show no signs of damage due to the period of profound shock. His mentality was

normal. There were no renal manifestations. The day after the operation, he was allowed to turn at will. He preferred lying on his back, but could turn on his right side. However, whenever he started to turn on his left side, for the first week or ten days, he felt "as if he were going to faint." This also happened whenever he raised himself suddenly. These symptoms were accompanied by the objective signs—pallor, increased pulse rate up to 120 and thready pulse, and a drop in his blood pressure to 90/60. Electrocardiographic study immediately after operation and during the convalescence was not particularly significant.

By the end of the third week after operation, he was well enough to leave the hospital; he could turn in any position he wanted to but still did not like to lie on his left side because it made him "feel funny."

Now six months after operation, he is doing well, he looks in as good health as before operation, he is out-of-doors, but does not yet feel able to go back to his job, which was one of physical labor.

SUMMARY

A patient was operated upon for a carcinoma of the left lung. The pneumonectomy included the removal of an adherent piece of parietal pericardium about 2½ inches in diameter overlying the region of the conus arteriosis. When the patient's position was shifted after the operation, the patient suddenly went into profound shock which he was able to tolerate for about 20 minutes, during which time means to combat the shock were tried. When the chest was re-opened, it was found that the heart had herniated through the pericardial defect and had become angulated and partially strangulated. As soon as the herniation was reduced, the symptoms disappeared. Recurrence was prevented by removing practically the entire anterior parietal pericardium. Recovery was complete and the removal of the pericardium seemed to produce no untoward effects.

UROLOGY AWARD

"*Urology Award*—The American Urological Association offers an annual award of $1000.00 (first prize of $500.00, second prize $300.00 and third prize $200.00) for essays on the result of some clinical or laboratory research in Urology. Competition shall be limited to urologists who have been in such specific practice for not more than five years and to residents in urology in recognized hospitals.

"The first prize essay will appear on the program of the forthcoming meeting of the American Urological Association, to be held at the Biltmore Hotel in Los Angeles, May 16–19, 1949."

For full particulars write the Secretary, Dr. Thomas D. Moore, 899 Madison Avenue, Memphis 3, Tennessee. Essays must be in his hands before February 15, 1949.

MYCETOMA — MADURA FOOT*

A CASE OF MYCETOMA PEDIS IN CHICAGO

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THE EARLIEST DESCRIPTION OF MYCETOMA was reported in the Madras Presidency of India in 1842 by Gill.[1] The condition had been known there for many years. In 1846 Colebrook from the same dispensary described further cases and introduced the term Madura foot for a distinct disease entity of that region. It was Vandyke Carter[2] who studied the condition over a period of years, 1860–1874, describing the changes caused in the bony structures and soft tissues of the affected parts and then identified filaments of a fungus found in the grains within the sinus tracts of the diseased area. He introduced the term Mycetoma, literally fungus tumor, for the condition, since he found it could affect other parts of the body than the foot. His pen and ink drawings and his descriptions indicate a true fungus with septate filaments and chlamydospores was studied. He classified his cases by the color of the grains found in the sinus tracts as pale or white, black and red.

Pinoy[3] in 1913 recognized the possibility of classifying cases of mycetoma by grouping the causative organisms, and in 1916 Chalmers and Archibald[4] reviewed the reported cases and published such a classification dividing them into two groups:

GROUP 1. Maduromycoses, caused by true fungi exhibiting septate filaments usually with chlamydospores, and
GROUP 2. Actinomycoses, due to delicate nonseptate filaments of the Actinomyces which belong to higher bacteria.

The basis for this classification has had to rest principally upon prepared sections from preserved material in the absence of cultures. After a critical examination of the first 25 cases of mycetoma recorded in Canada and the United States, Jones and Alden[5] in 1931 reported that 16 could not be definitely classified because of inadequate data. Seven cases were clearly of Maduromycoses and two were of Actinomycoses. In 1945, Burns, Moss and Brueck[6] published a chart covering the 38 cases reported up to April 1944. The chart which follows gives a similar picture of each case reported in this country from that date to and including the present case, thus bringing all recorded cases up to the present time.

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TABLE I.

Number of Case	Authors	Date Reported	Age at Report	Age Infected	Sex	Race	Occupation	Site of Infection	Where Acquired	Predisposing Trauma	Color of Grains	Classification of Organism	Cultures
1.	Symmers and Sporer[12]	May 1944	67	65	M	American (New York)	Clerk	Hand	New York	Splinters in hand (fall)	Black	Maduromycoses Chlamydospores	Negative[12]
2.	Venable and Gaston[13]	June 1944	8	3	M	Negro	Farmer's child	Left foot	Georgia	Glass in Foot (barefooted)	Yellow	Maduromycoses	Cultured and studies
3.	Gottlieb, A.[14]	June 1944	42	37	M	Mexican	Farmer	Left foot	California	Callus near heel	Black	Maduromycoses Clubs	Negative
4.	Gottlieb, A.[14]	June 1944	55	49	M	Mexican	Farmer	Left foot	California	Wore shoes; no socks	Black	Maduromycoses Septate hyphae and clubs	Negative
5.	Twining, Dixon, and Weidman[15]	June 1944	42	35	M	American Italian	Soldier in 1918	Right foot	Pennsylvania	Injury	Grey white	Maduromycoses Monosporium Apiospernum	Proved by culture[20]
6.	Twining, Dixon, and Weidman[15]	1944	42	40	M	Filipino	Sailor	Right foot	Alaska	Bruised by cable	Black	Maduromycoses Cephalosporium granulomatus	Proved by culture[20]
7.	Hatch and Wells[16]	August 1944	54	42	M	Finn	Railroad	Right foot	Minnesota	Fractured twice	?	Actinomycoses	Negative
8.	Clough, F. E.[17]	December 1944	53	43	M	Mexican	Farmer	Left foot	California	No history	Brown	Actinomycoses	Negative
9.	Wood, D. A.[18]	March 1945	40	39	M	Filipino	Sailor	Left foot	Treasure Island	No history	Black	Not identified Pickled	No cultures
10.	Peters, J. H.[19]	March 1945	53	51	M	Negro	Ditch digger	Left foot	Louisiana	Stood in water	Yellow	Actinomycoses Nocardia asteroides actinomyces	Proved by cultures
11.	Green, Bolton, and Woolsey	November 1946	35	19	M	Negro	Janitor	Right foot	Tennessee	Stepped on thorn	Ochroid	Actinomycoses Nocardia Madurae	Proved by cultures

In our review of the reported cases in this country, three were found to belong to Mycetoma Actinomycoses by the isolation of the causative filaments in pure culture and further study for their identification. The cases mentioned by Jones and Alden were those of Allison[7] in 1912, and of Lovejoy and Hammack[8] in 1920. Allison isolated an Actinomycete from his case in Texas of a Mexican gardener, and identified it as Streptothrix Madura. Lovejoy and Hammack sent their strain, recovered from a Mexican ranch laborer in California, to Boyd and Crutchfield,[9] who identified it a year later as Actinomyces Mexicana.

FIG. 1.—Marked enlargement of right foot.

In 1945, Peters[10] treated a case of Madura foot in New Orleans with sulfonamides and checked the progress of the treatment by cultures. Burns and Brueck isolated an Actinomycete from a case which they identified as Actinomyces asteroides. The case reported below represents only the fourth case in the United States which has been proven by cultures to belong to the Actinomyces group.

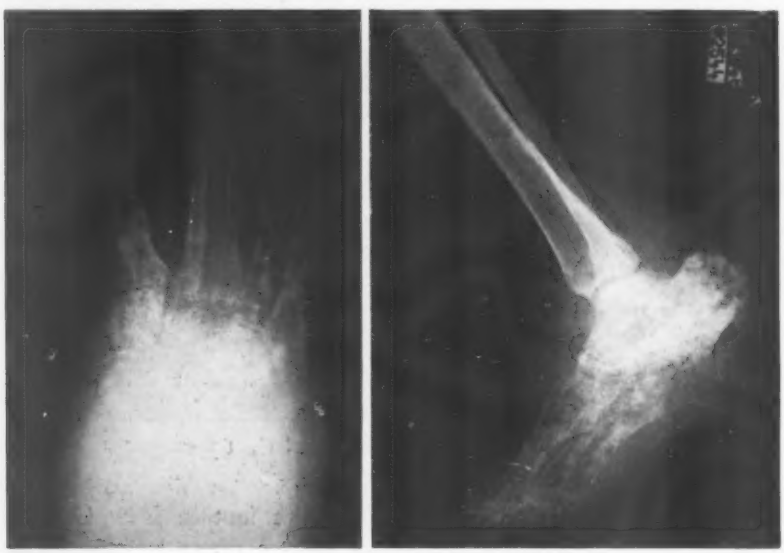
CASE HISTORY*

W. M., a 35-year-old colored male who had never been out of the United States, gave the following story. He lived in Memphis until late in 1944, at which time he moved to Chicago. He had never worked on a farm, but had done janitor work. The only antecedent history was that in 1925, while barefooted, he stepped on a thorn. Shortly thereafter the right foot became painful on motion, and he remained off his feet for 3 weeks. There was no swelling or suppuration, and he had no symptoms from 1925 to 1944, an interval of 19 years. Early in 1944, swelling of the foot appeared, and after persisting for several weeks, it subsided, and he was symptom free for 6 months. Swelling then again appeared, which subsided with rest, and about one year ago, a papule appeared on

* This case was presented before the Chicago Surgical Society on November 1, 1946.

the dorsum of the foot; the papule broke down and a serosanguinous drainage was present. This subsided, but other papules appeared, until a great number of papules with draining sinuses were present, along with gradual but marked enlargement of the foot (Fig. 1) until finally he presented a foot, which because of its size, offered mechanical disability. There have been, and are, no subjective symptoms such as pain, fever, or systemic manifestations.

Physical examination upon admission to the hospital demonstrated a foot substantially as illustrated, with the exception that there was a serous drainage from many of the now covered papules. Blood picture and chemistry, repeated complement fixation tests, chest roentgenograms, and roentgenograms of other bones were all within normal limits. Roentgen-ray examination of the foot (Fig. 2, a and b) showed extensive destruction. It revealed numerous circumscribed, punched-out areas of radiolucency with deformity of the shafts of all the bones, and with destruction of the joint spaces. There was marked soft tissue swelling.



A B

FIG. 2(A)—Extensive destruction of foot, as described in text.
(B)—Extensive destruction of foot. Tibia and fibula appear normal.

Bacteriologic studies. Smears and cultures were made. The grains used for smears and cultures were removed surgically from the bases of the raised, moist papules that formed part of the characteristic appearance of the foot. This procedure was repeated upon 3 consecutive occasions so that cultures were started at intervals on Sabouraud's medium, both dextrose and maltose, and on Herrold egg yolk agar.[11]

The grains were pale or ochroid in color, hard and brittle. Smears, made by crushing and rubbing a grain between microscope slides, consisted of delicate nonseptate filaments less than 0.5 μ in diameter, and showed true branching (Fig. 3). The filaments were Gram negative containing Gram positive granules. They were not acid-fast. Grains were seen up to 3 mm. in diameter.

From none of the original cultures planted on the Sabouraud's media was growth of any kind recovered. However all the Herrold egg yolk agar cultures produced growth in varying degree by the tenth day. The colonies were tiny and brittle and all smears

FIG. 3

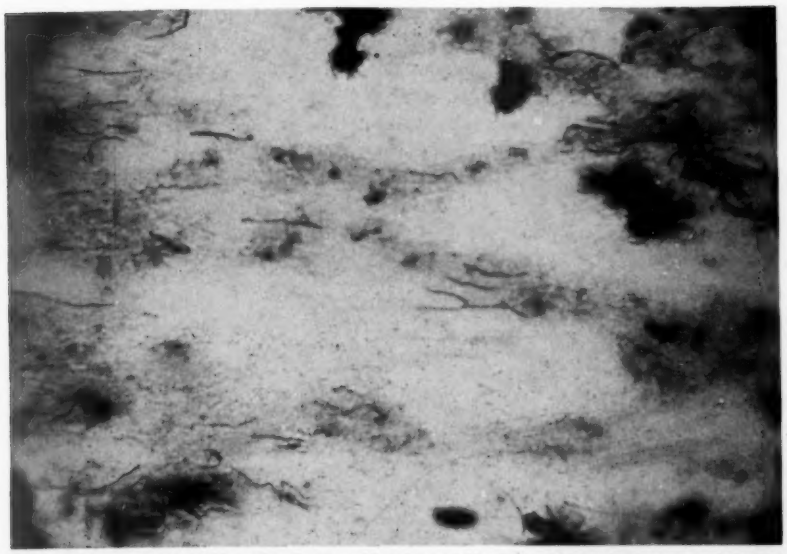


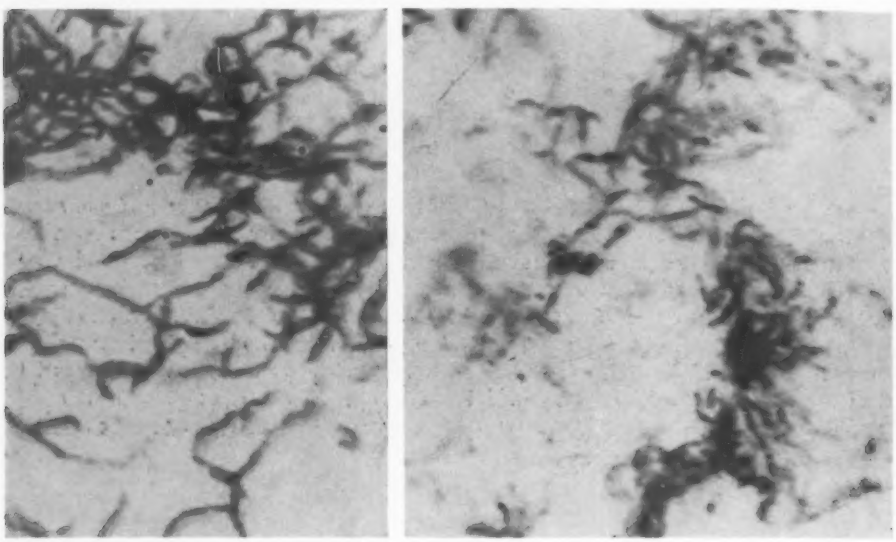
FIG. 4

FIG. 3.—Photomicrograph showing contents of a crushed grain from the sinus tract of the foot. Filaments were under 0.5 μ in diameter.

FIG. 4.—Filaments of the same organism from a culture. Slender pointed Gram negative tips are numerous in the periphery of the cluster and Gram positive granules are noticeable in the older part of the filaments.

showed the same type of filament found in the original grain. The outstanding characteristics of these filaments from cultures were the long pointed ends definitely Gram negative without Gram positive granules near the tips (Fig. 4). They were under 1 μ in diameter. Unusual forms were not found. In the tangle of the older filaments Gram positive granules were very noticeable. True branching was found occasionally.

The organism grew aerobically equally well at room and incubator temperatures. By subculturing the luxuriant growth from the Herrold egg yolk medium, new colonies were recovered in 6 days on blood agar, chocolate proteose agar, Loeffler's blood serum, and Herrold's egg yolk agar. Loeffler's medium was at no time liquefied. Growth was



A B

FIG. 5(A.)—Gram positive club-like structures at the ends of faintly staining filaments. Special culture.
(B.)—Gram positive diphtheroids as found in many fields. Special culture.

recovered in plain broth as small pellets in the bottom and adherent to the sides of the tubes. In brain broth the pellets adhered to the surface of the pieces of brain. The broth did not become cloudy and filaments were not found in the smears. It was after the strain had been subcultured repeatedly on the egg yolk agar that generous transplants produced growth on both the dextrose and maltose Sabouraud's media.

The typical colonies were smooth, rounded and hard (Fig. 6), and did not produce aerial hyphae. They were hollow and grew tenaciously into the medium. Usually an entire colony or cluster of colonies was transferred for subcultures since they were too hard to break with a platinum wire loop. Occasionally a few tiny white flecks of aerial hyphae were noted on very old cultures where the medium had become paper thin. This hyphae consisted of strongly Gram positive filaments 1.5 μ in diameter and varied in length, some reaching 10 μ. No unusual forms were seen.

The ochroid color of the original grains prevailed in the majority of cultures on all media tried. However, on the Herrold egg yolk medium at times the young ochroid colonies soon turned into rich shades of coral, orange, or raspberry. Eventually young growth on Sabouraud's and Loeffler's media acquired the coral color. When the coral colored growth aged, it became a definite raspberry shade or a deep red. These colors were kept in stock by subculturing them on egg yolk agar, but they often reverted to the ochroid shade. At no time was the color absorbed by the medium.

While studying the colored colonies, smears and cultures were made from spreadable material obtained from the lining of one of the ball-like colonies of young coral growth on the Herrold medium. These cultures proved to be no different from others and the organism was still non acid-fast, but the long pointed tipped filaments (Fig. 4) were absent. Now, heavily Gram positive diphtheroids (Fig. 5b) were prominent as well as faintly staining filaments ending in Gram positive swellings or club-like structures (Fig. 5a). A few branched filaments were noted. The diphtheroids ranged in length from 2 μ to 7 μ by 0.7 μ in diameter, while the club-like ends averaged 1 μ x 7 μ and varied in shape.

From the work accomplished to date, the organism isolated from this case of Madura foot belongs to the Actinomycoses group and is a strain of Nocardia Madurae. Rabbits injected nine months ago with a suspension of the growth remained healthy. Further cultures are contemplated and an

FIG. 6.—Ochroid colonies of the organism as it grows on Herrold Egg Yolk Agar.

attempt is in progress to find an explanation for the extreme hardness of the growth and for the source and variable appearance of the colors. We hope to submit a report upon these results at a later date. Our findings have been confirmed by Dr. C. W. Emmons, Principal Mycologist, National Institute of Health, Bethesda, Maryland.

On the basis of Peters' experience, previously mentioned, our patient received what we believed to be adequate amounts of sulfadiazine. Although the foot seems substantially free from infection, yet there has been no appreciable reduction in its size. Because of its bulk, it presents a mechanical problem to the patient, and it is felt that amputation will be resorted to.

It is possible that this entity may appear in the future among war veterans who will have been exposed to the etiological agents, and thus it is important to keep this condition in mind. It is probably most frequently confused with chronic osteomyelitis, chronic cellulitis, tuberculosis, and syphilis. It is possible that this condition is more prevalent than is generally assumed.

SUMMARY

A case of Mycetoma-Madura Foot is described, proven by cultures, and representing, according to the literature reviewed, the fourth case in the United States which has been so proven. This actinomyces type is in contradistinction

to the more common type which is a true fungus. We have isolated the organism in this case in pure culture and have grown it on a variety of media, including Herrold's egg medium.

Madura Foot is a granulomatous condition resulting in marked deformity and enlargement of the foot, with destruction of the internal structures. There are nodules on the external surface which connect with granulomatous lesions of the interior of the foot by sinuses. These sinuses discharge granules of various sizes and colors, and it is from these granules that the organism is recovered.

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VOLVULUS OF THE SIGMOID COLON*

Report of Two Cases

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DUE TO THE RELATIVE PAUCITY of the reports in the American literature regarding volvulus of the sigmoid and the distressing lack of information in the usual reference books on the subject, we feel it pertinent at this time to give a brief discussion of the condition and to present two recent cases.

An interesting observation made while reviewing the literature was the fact that to 20 reports found in the American, British, and Australian literature 85 reports relative to volvulus of the sigmoid were found in non-English literature. A possible explanation for this fact is that the condition occurs with considerably more frequency in Russian and Baltic people than in British and American people.[9] It is of relatively common occurrence in Germany where it is reported to be the cause of large bowel obstruction in as high as 30 per cent of the cases. The percentages in this country are much less, but vary considerably. The condition is reported by Hinton and Steiner[7] to account for less than one per cent of acute intestinal obstruction and by Griffin, Bartron and Meyer[13] to be the cause of eight per cent of a series of 458 cases of large bowel obstruction at Cook County Hospital. Miller[3] reports it as the cause of 15 per cent of all forms of acute intestinal obstruction. (These are all American case figures.) An explanation for this inconsistency in percentages is perhaps the relative rarity of the condition. Few have been able personally to observe more than a small number of cases at most. An interesting explanation for the higher incidence of the condition in some other sections of the world, Russia and the Baltic area—for instance, is the dietary habits of these peoples. They seem to be more of vegetarians than we Americans, and their diet, consisting of coarser foods containing much cellulose, makes a greater demand upon the pelvic colon as a storehouse.[9] This might be conducive to a tendency toward redundance of that part of the bowel.

ETIOLOGY

There is general agreement that an enlarged colon and a redundant mesentery definitely predispose to volvulus; the former is usually present, and the latter invariably is found. A narrow attachment of the mesentery is listed by Hinton and Steiner[7] as a predisposing factor, but it seems that this is most likely a relative condition; since the attachment is a stable affair and cannot elongate commensurately with the mesenteric reflection at the bowel. Ligot and Overend[9] state that there is a familial tendency to congenital elongation of the pelvic colon and, hence, a predisposition to volvulus. There is

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general agreement that a history of chronic constipation is the rule. Heavy work and drastic purgation are suggested as precipitating causes.[9]

Volvulus of the pelvic colon is a disease of the older age group, most of the cases occurring in patients past the age of 40 years. It can occur in children—however, and Miller[3] reports a case occurring in a child 12 years old. Our two cases were 61 and 70 years of age respectively.

CLINICAL PICTURE

The history is that of acute low bowel obstruction. These patients usually are seen by a physician after one to several days of symptoms of partial or complete low bowel obstruction. The possible explanation for this is that it is the rule for these patients to have had several previous episodes of a similar nature which they have been able to relieve by means of purgatives and/or enemas without consulting a physician. This observation is substantiated by reports in the literature. Ligot and Overend[9] stress the importance of a history of diarrhea *after* the abatement of the cramping abdominal pains rather than *during* the pains. A history of chronic constipation is suggestive. It would be superfluous to reiterate the physical findings as regards the abdomen since they are those of acute low bowel obstruction from any cause.

The fluoroscope and roentgen-ray are important adjuncts in the diagnosis. Hall[4] lists three findings as his criteria for making a roentgen-ray diagnosis of this condition: (1) a tremendously dilated sigmoid colon situated predominantly in the right side of the abdomen, (2) valve-like obstruction distal to the dilation, allowing free entry of the enema fluid, yet preventing the patient from evacuating the contents from the dilated loop, and (3) normal mucosal pattern in the sigmoid distal to the dilated loop. Griffin, Bartron, and Meyer[13] found that 76 per cent of their patients were unable to take more than 500 cc. of enema fluid. A flat plate of the abdomen is of assistance when one observes a tremendously dilated loop of bowel rising out of the pelvis, frequently up as high as the liver, in the form of an inverted "U".[9]

A long history of repeated attacks is helpful in differentiating this condition from obstruction due to carcinoma, which runs a more progressive course.

TREATMENT

Once the diagnosis is made, the treatment is surgical—after the patient is properly prepared as regards fluid and electrolyte balance. Conservative surgery, in the form of fixation of the redundant bowel to the abdominal wall, was unsatisfactory in our hands as regards permanent relief of symptoms. This view is substantiated in the literature[11] by others who have performed this same conservative procedure. Likewise, mesentery-shortening procedures have been tried with the same poor results, and surgeons have been chagrined, when operating upon these patients for a recurrence following these conservative measures, to find no evidence of their painstaking efforts during the previous operation. This was our experience. It seems to be the consensus of opinion, and is our firm conviction, that the only rational treatment of

volvulus of the sigmoid is resection of the redundant bowel, preferably a Mikulicz resection. The lower mortality in the literature has been in the cases upon whom a Mikulicz resection was performed. Weeks[2] reports in a review of the literature up until 1931, 65 cases attended by a 33⅓ per cent mortality. Eighteen of these cases had a three-stage Mikulicz operation with but two deaths. Our experience was that the procedure is no more shocking than the more conservative surgery. Although occasional good results are reported in the literature of patients upon whom resection and primary anastomosis was performed,[17, 16, 12] we feel that the Mikulicz resection is less likely to be attended by complications such as might be experienced with primary anastomosis; in addition, the technical difficulty of uniting the dilated upper segment of the descending colon to the smaller distal portion sigmoid is great, considering the poor condition of the dilated bowel wall.

Case 1. No. 4843: White male 61 years of age admitted January 29, 1942, complaining of cramping abdominal pain, vomiting, and inability to pass flatus or feces. He remembered having a similar attack about a year previously, which abated spontaneously after 24 hours.

The examination was essentially negative except for a markedly distended abdomen with visible peristalsis. There was no muscle spasm. At this time a diagnosis of intestinal obstruction was made, and decompression by means of a Wangensteen suction was begun. The distention was not relieved by the next day at which time a barium enema was attempted. The barium was found to meet an obstruction after passing upward about six inches. During this procedure the obstruction was partially relieved, and a large quantity of liquid fecal material escaped, completely relieving the distention. Two days later the barium enema was repeated, and a partial obstruction was found at the recto-sigmoid junction. No growth could be visualized through the proctoscope.

On February 4, 1942, an exploratory laparotomy was performed, and no abnormalities of the abdominal viscera could be found other than a nodular liver and a somewhat dilated sigmoid with very redundant mesentery. It was estimated that the sigmoid could be elevated 12 to 16 inches outside the abdomen through the incision. The incision was closed with silk. The recovery was uneventful, and the patient was discharged on the tenth postoperative day, asymptomatic.

Second Admission.—Admitted February 26, 1945, with symptoms of low bowel obstruction. He was operated upon shortly after admission through a low, left paramedian incision, and a volvulus of the sigmoid was found. The volvulus was relieved manually, and the redundant sigmoid sutured to the abdominal wall on both sides of the incision with fine, interrupted cotton sutures. The wound was closed with interrupted cotton sutures. The patient was discharged on the seventh postoperative day.

Third Admission.—Admitted December 13, 1946, with symptoms of low intestinal obstruction of 18 hours duration. Two hours after admission the obstruction relieved itself spontaneously, and the patient was completely relieved of symptoms and began to pass flatus and feces. He was discharged asymptomatic the following day.

Fourth Admission.—He again reported on February 23, 1947, to the hospital with symptoms and signs of low bowel obstruction of 48 hours duration. After a short period of conservative efforts, with failure to relieve the obstruction, he was operated upon. This time a tremendously distended sigmoid colon was found, which was twisted upon itself and completely obstructed. No evidence of previous suturing of the bowel to abdominal wall was found. A Mikulicz resection was performed, about 18 inches of the sigmoid being thus resected. The incision was closed with interrupted cotton sutures. Two days later the clamps were removed from the ends of the divided bowel, and four

days after the clamps were removed (sixth postoperative day) a crushing clamp was applied.

Thirty-one days after the resection was performed the colostomy was closed. The skin sutures were removed seven days later, and the patient was discharged with normal bowel movements and no drainage from the wound.

Case 2. No. 7613: White male 70 years of age admitted to this hospital on December 6, 1946, complaining of abdominal distention of about 15 hours duration and inability to pass flatus or feces, despite purgatives and enemas. There was no nausea or vomiting. He gave a history of several similar episodes within the previous three years, which were relieved by enemas, but about which he had not consulted a physician. The remainder of the history was essentially non-contributory. An appendectomy was performed upon him in 1924.

The examination revealed a thin, white male appearing to be about the stated age with a very dry mouth and tongue. There were scattered moist rales over the base of the left lung, a markedly distended abdomen. The colon could be visualized throughout its course, and increased peristalsis could be auscultated throughout. No definite masses or localized tenderness were found. Rectal examination revealed no masses or obstruction within reach of the finger. The roentgen-ray of a barium enema showed the barium to stop about six inches up in the rectum, the blind end being of smooth outline. The diagnosis at this time was low intestinal obstruction, cause undetermined, and he was prepared for surgery.

Through a low, left rectus incision the abdominal cavity was opened, and an enormously distended sigmoid was found as the result of volvulus, which—when the torsion was relieved—was found to be quite atonic with an unusually long mesentery. The redundant sigmoid was sutured to the left lateral abdominal wall with interrupted sutures of fine cotton and the wound closed with interrupted cotton sutures. The patient was discharged on the fifteenth postoperative day in good condition.

Second Admission.—Since being discharged from the hospital about two and one-half months before, the patient had enjoyed good health until 24 hours before admission. He began to experience cramping abdominal pains, to notice that he was becoming distended, and was unable to pass flatus or feces.

The physical examination was essentially the same as on the previous admission. He was again prepared for surgery.

The operative findings were essentially the same as on the previous occasion with no evidence of the colon having been sutured to the abdominal wall. The volvulus was relieved and a Mikulicz resection performed. The clamps were removed from the cut ends of the bowel two days later and a spur clamp applied on the sixth postoperative day.

Four weeks postoperatively the colostomy was closed. His subsequent course was uneventful except for a fecal fistula at the site of the closure which closed spontaneously before his discharge on the fifty-first postoperative day.

This patient has remained asymptomatic.

COMMENT

Judging from the reports in the literature volvulus of the sigmoid is of relatively infrequent occurrence, but it is common enough that every surgeon should familiarize himself with the pros and cons as regards conservative and more radical surgery. From our experience with these two very similar cases and from the information gleaned from the literature, we are of the opinion that a resection of the Mikulicz type at the initial operation is the operation of choice and that it is very little—if any—more shocking than the more conservative procedures. In each of these cases the patient might have been spared the added expense and inconvenienc of mor than one admission for the

same condition, had this view been taken at first. Of course, the follow-up is not of sufficient duration for one to make any conclusive statements as regards the permanency of the present freedom of symptoms, but—anatomically—one would expect it to be more successful than the more conservative surgery which was in our hands.

SUMMARY

A brief discussion of volvulus of the sigmoid colon is given. Two cases are presented of recurrent volvulus of the sigmoid following conservative surgery, which subsequently required partial sigmoidectomy for the relief of symptoms.

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GASTRODUODENAL INTUSSUSCEPTION*†

A CASE REPORT

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SIMPLE PROLAPSE of the antral mucosa through the pylorus, with or without associated gastric polyps is quite common, but actual intussusception of all coats of the stomach wall is very rare.

Intussusception of the degree encountered in the case described below is almost a medical curiosity.

CASE REPORT

H. N., No. 44987. The patient was a dental assistant, 35 years of age, who was admitted to the Roosevelt Hospital for the first time on July 29, 1947. She complained of well localized epigastric distress of about three months duration. The initial symptom had been a puzzling and persistent eructation of gas and sour liquid after meals, associated with a feeling of distention and pressure in the upper abdomen. As the weeks passed, despite numerous modifications in her diet, the discomfort became more accentuated. During the early days of her illness, small doses of bicarbonate had given some relief, but after a while it induced vomiting and the patient discontinued its use. At the time of entry into the hospital, she was nauseated most of the time. Although there had been no lack of appetite, she had practically stopped eating because food immediately aggravated the discomfort and was promptly regurgitated. During the preceding two months she had lost 12 pounds in weight. No gross bleeding had been observed and the patient had not noticed any discoloration of the vomitus or of the stools.

Her past history failed to disclose any previous attacks of a similar nature. As a matter of fact, she had never been disturbed by gastro-intestinal symptoms of any sort except for a very rare attack of diarrhea.

On physical examination there had been obvious weight loss. Blood pressure, pulse, respirations and temperature were normal. There was a distinct pallor but no jaundice or cyanosis. No cervical lymph nodes were palpable. The heart and lungs were entirely normal. Examination of the abdomen revealed only slight mid-epigastric tenderness and a sense of fullness in the mid-epigastrium without any definite mass. The extremities and reflexes were normal.

Within a few days after admission a gastro-intestinal series was done (Figs. 1 and 2) and the following findings were noted. The esophagus was normal. There was a large gastric defect involving the fundus and antrum with distortion of the duodenal bulb. There was lack of filling of the descending duodenum without any delay, however, in the emptying time of the stomach.

Lateral views showed essentially the same picture, with what appeared to be forward displacement of the stomach and antrum.

The diagnosis lay between a large benign intragastric tumor or a large primary tumor of the pancreas producing a pressure defect in the antrum and fundus. A gastroscopic examination was not permitted by the patient.

Following additional clinical studies, all of which failed to provide any further diagnostic assistance, operation was performed on July 14, 1947.

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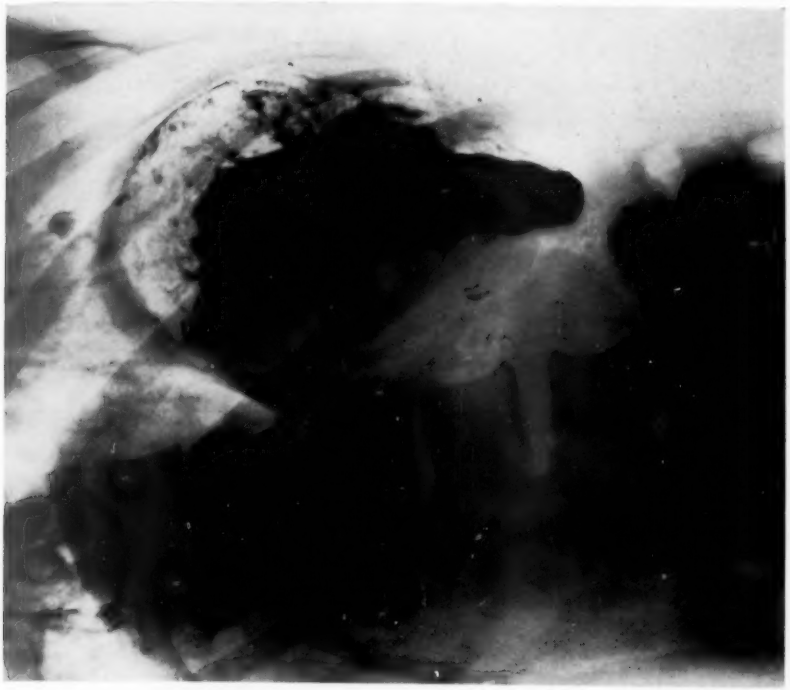

FIG. 2.—Lateral oblique view.

FIG. 1.—Antero-posterior view of stomach and duodenum following ingestion of barium.

FIG. 3.—Gastroduodenal intussusception as found at operation.

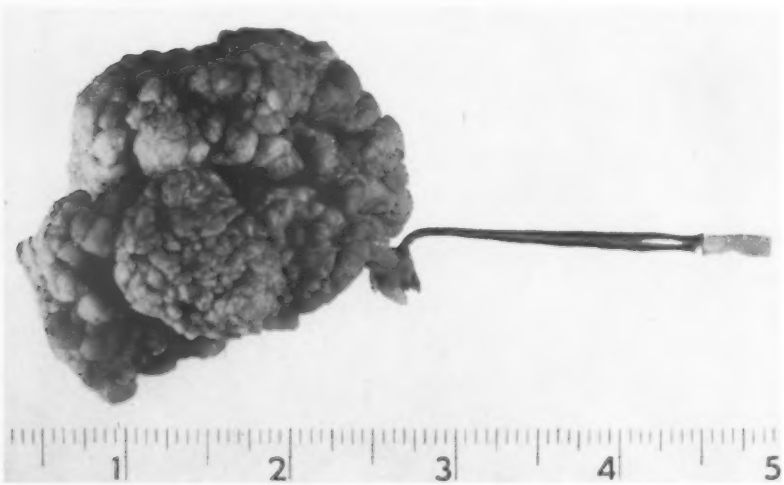

FIG. 4.—Gastric polyp removed at operation. The hook indicates the pedicle of the tumor.

The situation found at operation is illustrated in Figure 3. About two thirds of the stomach had herniated through the pylorus into the duodenum. As a result of this intussusception, the duodenum was dilated to about three inches in diameter down to the region of the duodenojejunal flexure. Although the pyloric muscle was still fairly tight, the intussusception was reduced without too much difficulty. As the stomach was delivered through the pylorus, a soft tumor was felt springing up through the pyloric ring as though attached to a large rubber band. Palpation of the stomach then revealed a mass about the size of a billiard ball within the gastric cavity. A long

FIG. 5.—Microphotograph of one of many sections made through various parts of the tumor.

gastrotomy was performed and a soft spongy tumor was found attached by a pedicle one inch long to the upper posterior third of the stomach wall. Since the tumor looked and felt benign, and the pedicle was obviously not involved, a simple removal of the tumor was performed by elliptical incision around the base of the pedicle. The resulting defect was closed with interrupted sutures of fine silk. Inspection of the gastric cavity failed to reveal any other polyps. The gastrotomy was then closed with two layers of interrupted Halsted sutures. The abdominal wound was closed in layers.

Figure 4 shows the tumor as removed, and Figure 5 is a microphotograph of one of the many sections from various portions of the tumor.

Microscopic examination showed simple benign adenomatous polyp.

The patient made a smooth recovery and has been entirely free of symptoms since operation.

CHOLECYSTITIS DUE TO GIARDIA LAMBLIA IN A LEFT-SIDED GALLBLADDER*

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THERE HAVE ONLY BEEN seven reported cases of infection of the gallbladder due to giardia lamblia in the literature to date. However, there is increasing evidence that this protozoa is the causative factor in cholecystitis more frequently than the rarity of reports would indicate. The case herein reported was one in which giardia were proven to be the cause of infection in a gallbladder. It is all the more unusual since the gallbladder was found to be situated on the left side. The giardia lambia was found repeatedly on duodenal drainage. The organism promptly disappeared under treatment with atabrine. The cystic form was recovered from the gallbladder after surgical removal. This is the eighth reported case of giardiasis of the gallbladder, and probably the second in which cysts were found in the gallbladder.

Leuwenhoek[10] first discovered giardia in 1681, in a specimen of his own stool. The organism was rediscovered and named by Lamb[5] in 1859. It is a protozoa measuring 8 to 20 mu. long and 5 to 12 mu. wide. In other words, it is larger than a red blood corpuscle, and smaller than an epithelial cell. It contains four ventrally placed flagellates, and is actively motile. It is a common inhabitant of the gastro-intestinal tract, and was formerly considered non-pathogenic. Its pathogenocity, particularly in the case of diarrhea, has been definitely established. Berberian[1] reviewed the reports of 15 authors on the pathogenocity of giardia lamblia with reference to diarrhea. Several of these authors definitely established it as the causative organism in epidemics of diarrhea, particularly in the Middle East. Routine examination of stool specimens in this country and abroad showed that the highest incident of giardia lamblia occurred in areas where sanitation was at its lowest. In Puerto Rico, 48 per cent of the population were found to be infested. A study of Chicago dispensary patients revealed a rate of only 1.66 per cent; while in Tenessee the rate was much higher at 14.7 per cent.

While proven cases of infection of the upper gastro-intestinal tract with this parasite have not been so common as those in connection with diarrhea, there is increasing evidence that the organism produces gallbladder disease,

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and probably more frequently than has been supposed. Smithies[11] claimed to be the first to report a case of giardiasis of the gallbladder. He found the protozoa in a gallbladder which had been removed by the late Dr. A. J. Ochsner of Chicago in December, 1917. He later reported a second case.

Keyes[9] reported two cases of cholecystitis, in one of which he found cysts of giardia in the gallbladder. Hartman,[8] in 1942, in a review of the literature, found reports of only four cases of giardiasis of the gallbladder. He added a case of his own, however, he omitted the two cases of Keyes. Search of the literature since that date has failed to reveal any reports of proven cases. Reported cases to date of cholecystitis due to giardia lamblia may be summarized as follows:

Smithies	2 cases
Westphal and Gerogi[13]	1 case
Calder and Ridgon[3]	1 case (autopsy)
Hartman	1 case
Keyes	2 cases
Total	7 cases

Another indication of the pathogenocity of the giardia lamblia in relation to the upper gastro-intestinal tract is the increasing number of reports in the literature showing the frequency with which it is found in duodenal drainage in association with symptoms simulating disease of the gallbladder or duodenum. Wesselman[12] reported two cases presenting symptoms simulating cholecystitis in which giardi were found on duodenal drainage. Prompt and complete relief was obtained by administration of acranil. DeMuro[4] reported a series of 45 cases in which he recovered giardia from the bile obtained on duodenal drainage. Of these, 31.1 per cent suffered from upper gastro-intestinal symptoms which he referred to as "entero-hepato-biliary syndrome." Goss[5] examined the stools of 300 patients for giardia, and found this protozoa to be present in 9 per cent. However, more interesting is the fact that in those suffering from symptoms simulating ulcer or gallbladder disease, the instance was 34 per cent. He believes that giardiasis leads to vitamin deficiency.

For the treatment of giardiasis, atabrine is a specific; although good results have been reported with other forms of therapy. Boros[2] treated a case of giardiasis of the duodenum simulating ulcer with five injections of 0.45 Gm. neosalvarsan at five-day intervals with good results. Goss[5] recommends, among other things, carbarsone. Berberian[1] found acranil 100 per cent effective in eliminating this infection in cases of stool contamination. The recommended dosage is as follows:

Age 3–6	0.1 Gm. per day for five consecutive days
7–12	0.1 Gm. twice daily for five consecutive days
13–16	0.1 Gm. three times daily for five consecutive days
Over 16	0.5 Gm. per day for the first day, and then 0.1 Gm. three times daily for four more days.

Hartman[8] and associates obtained a cure in their case of giardiasis of the gallbladder with atabrine in doses of 0.1 Gm. three times daily for five days.

Gross[6] reviewed the literature on *gallbladder anomalies* thoroughly in 1936, and was able to find only seven cases of left-sided gallbladder. In each case the vesicle was situated in the under surface of the left lobe of the liver. The gallbladder was usually found to be normal in size. The cystic duct joined the common hepatic duct in the normal manner and position in three cases, the left hepatic duct in one and unrecorded in three. The embryologic development of this anomaly may occur in one of two ways. The gallbladder anlage begins as a normal embryologic bud from the hepatic diverticulum and migrates to the left where it becomes fixed by the developing peritoneum to the under surface of the left lobe of the liver. This accounts for the normal entrance of the cystic duct into the hepatic duct. This was the situation in cases described by Harris and Walton.[7] They described the cystic duct as coming off at the normal site extending forward and to the right in the direction of the normal site of the gallbladder and then making a sharp hairpin turn to join the fundus under the left liver lobe. The second way in which this anomaly may develop is as follows: A gallbladder develops on each side with one persisting on the left and the right-sided one becoming atrophic and disappearing. In such a case one would expect the cystic duct to drain into the left hepatic duct as has been reported in one case.

Case Report

W. E. S., a white male, age 18, was admitted to Regional Hospital, Camp Joseph T. Robinson, on February 15, 1946. His *chief complaint* was pain in the right side of the abdomen of three days' duration. His *past history* was irrelevant, except for an appendectomy in 1944. He had not seen tropical service.

The history of the present illness revealed that three days prior to admission he developed pain in the right upper quadrant of the abdomen in the subcostal region. Pains had been sharp enough to "shut his wind off." Pain radiated to the left side, but not to the shoulder or directly to the back. He vomited twice. Vomitus contained food which he had previously eaten. Stools were normal in appearance and contained no evidence of blood. There was no history of intolerance to food.

Physical examination revealed a white male in a good state of nutrition. The temperature was 98; pulse 84; and respiration, 22. The abdomen was tender on palpation in the right upper quadrant. The liver was not palpable. The blood pressure was systolic 110, diastolic 60. Physical examination was otherwise negative.

Laboratory Tests and Special Examinations: Routine blood tests and urinalysis were within normal limits. The serum protein was 7.5; blood cholesterol, 1.36; and the blood Kahn was negative. The sedimentation rate was 7; hematocrit, 44; icteric index, 10.2. Examination of the bile obtained on duodenal drainage on March 8, 1946, showed *numerous giardia lamblia* present. These protozoa were clumped together in small rod-shaped structures, as if they had formed casts of the small radicles of the biliary tree. They were packed solid in these casts and occasionally one would break away from the general mass and swim in the fluid media.

X-ray examination of the gallbladder (Graham series) taken on February 20, 1946, showed no evidence of filling. Both kidneys appeared normal, and intravenous pyelograms were within normal limits. Roentgen examination of the upper gastro-intestinal tract failed to reveal any other pathology.

Course in Hospital: On March 7, 1946, the patient was put on daily duodenal drainage. Because of the repeated finding of giardia in the bile, administration of atabrine was begun, on March 16, 1946, in doses of 0.1 Gm. three times a day. This therapy was

FIG. 1.—This is a drawing from the microscopic examination of the bile obtained on duodenal drainage. In the upper left-hand corner is a mucous cast containing numerous giardia. This cast probably came from one of the small radicles of the biliary tree. In the lower right-hand corner is a high power of the organisms.

continued for one week at the end of which time the giardia had disappeared from the duodenal drainage. None were found subsequently from repeated drainages taken almost daily between March 26 and May 9. A second roentgenogram of the gallbladder showed no evidence of filling with the contrast media (Priodax). The cephalin flocculation test on March 5 was as follows: 24 hours plus and minus; 48 hours plus and minus. Examination of the feces on three successive days, starting on March 25, showed no evidence of parasites. The patient was operated upon April 5, 1946, at which time the gallbladder was removed and the common bile duct explored and drained.

The operative findings were as follows: Gallbladder was situated under the left lobe of the liver. The fundus was the same distance to the left of the falciform ligament as the

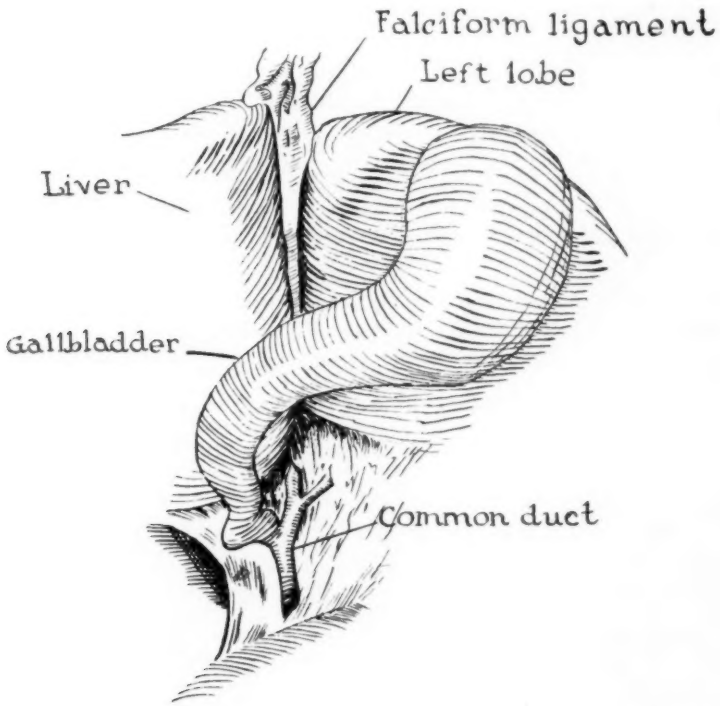

FIG. 2.—This is a diagram of the anomalous gallbladder in which the fundus was situated in the left lobe of the liver. The neck of the gallbladder crossed over to the right crossing the longitudinal fissure near its posterior end. The cystic duct joined the common duct on the right side.

normal gallbladder is usually to the right. The neck of the gallbladder crossed over to the right, crossing the longitudinal fissure near its posterior end. The cystic duct joined the common hepatic duct on the right side. The common bile duct, pancreas and other organs were essentially normal. The liver showed evidence of considerable hepatitis. It was moderately swollen and there were visible areas of fibrosis. These areas of fibrosis radiated over the surface of the liver substance from the gallbladder. There was further evidence of cholecystitis in the form of edema about the cystic duct and adhesions to the duodenum. No stones were found in the gallbladder or common bile duct. A T-tube was placed in the common bile duct for the purpose of subsequent study of the bile for evidence of giardia infection.

Pathologic Examination of scrapings of the gallbladder mucosa showed the presence

of giardia cysts. There was also evidence of ulceration in the mucosa of the gallbladder extending into the submucosa.

Postoperative Course: Patient made an uneventful convalescence. Repeated examinations of T-tube bile and also of duodenal drainage postoperatively were negative for giardia. The T-tube was removed at the end of six weeks. Patient has since been symptom free. Cholangiograms taken on May 1, 1946, showed the common duct and biliary tract to be normal.

Comment. This is a case of a man with left-sided gallbladder with typical symptoms of cholecystitis in whom giardia lamblia were found in large quantities in the bile obtained on duodenal drainage. These protozoa promptly disappeared following administration of atabrine as did the symptoms. Besides roentgen evidence, the gallbladder showed signs of inflammation at the operating table and giardia cysts were found in the gallbladder wall after surgical removal.

DISCUSSION

The evidence in favor of this being a giardia infection rather than an infestation is: (1) typical symptoms and physical signs of cholecystitis relieved by atabrine; (2) failure of the gallbladder to cast a shadow on the x-ray film after a Graham series on two occasions both before and after repeated duodenal drainages; (3) findings at the time of operation of streaks of fibrosis on the liver surface extending from the gallbladder and inflammatory reaction around the cystic duct; (4) pathologic study of the gallbladder showing ulceration in the mucosa and submucosa and the finding of giardia lamblia cysts in the scrapings of the mucosa.

SUMMARY AND CONCLUSIONS

1. While giardia lamblia in the past has been considered harmless there is increasing evidence that it is capable of producing gastro-intestinal disease.

2. Only seven proven cases of gallbladder disease from giardia have been previously reported in the literature; however, there have been instances of cases with symptoms simulating gallbladder disease or duodenal ulcer in which giardia lamblia were found in the bile obtained by duodenal drainage. Alleviation of symptoms was obtained by elimination of the protozoa by therapy.

3. Although various forms of therapy have been recommended, atabrine appears to be the drug of choice. The recommended adult dose is 0.1 Gm. three times daily for five days.

4. A case of giardiasis in a patient with a left-sided gallbladder is herein reported. In this instance the giardia were found in the bile obtained by duodenal drainage. They were eliminated by administration of atabrine and symptoms promptly subsided. When the gallbladder was subsequently removed, giardia cysts were found in the gallbladder wall.

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STATEMENT OF THE OWNERSHIP, MANAGEMENT, CIRCULATION, ETC., REQUIRED BY THE ACTS OF CONGRESS OF AUGUST 24, 1912, AND MARCH 3, 1933
Of ANNALS OF SURGERY, published monthly at Philadelphia, Pa., as of November 1, 1948.

State of Pennsylvania }
County of Philadelphia } *ss.*

Before me, a Notary Public in and for the State and county aforesaid, personally appeared J. R. Arnold, who, having been duly sworn according to law, deposes and says that he is the Treasurer of the ANNALS OF SURGERY and that the following is, to the best of his knowledge and belief, a true statement of the ownership, management (and if a daily paper, the circulation), etc., of the aforesaid publication for the date shown in the above caption, required by the Act of August 24, 1912, as amended by the Act of March 3, 1933, embodied in section 537, Postal Laws and Regulations, printed on the reverse of this form, to wit:

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[Signed] J. R. ARNOLD.

Affirmed to and subscribed before me this 7th day of October, 1948.
[Seal] HARRY J. BEARD.

(My commission expires March 5, 1949.)

MEDIASTINAL LIPOMATA: A CASE REPORT*

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Less than 50 intrathoracic lipomas have been reported. Seventeen of these have been successfully excised.[1, 2, 3, 4, 5, 6] Heuer and Andrus, in the first comprehensive report[1] in 1933, suggested three anatomic groups: the hour-glass tumors, in which part of the tumor lay within the thorax and the remainder penetrated the intercostal structures to present subcutaneously; the superior mediastinal lipomas which present at the root of the neck; and the entirely intrathoracic lipomas. The majority occupy the mediastinum. The disease is a rare clinical entity and therefore the following case is considered worthy of report.

CASE REPORT

E. M., a 30-year-old negress, had a routine chest roentgenogram in 1945 which showed a superior mediastinal mass. She was asymptomatic and did not seek medical aid. Another roentgenogram in 1947 revealed the mass with moderate increase in its size although she was still asymptomatic. She received four radiation treatments without any reduction in the size of the mass. The patient was then referred to one of us (E. H. G.) who advised surgery.

She entered the Methodist Hospital, Brooklyn, on November 3, 1947. History and physical examination were completely negative. Laboratory studies were negative with the exception of roentgenograms of the chest (Fig. 1) which showed an ovoid tumor mass anteriorly in the superior mediastinum, considered to be a dermoid or a lipoma.

On November 5, 1947, anterior thoracotomy was performed by the senior author through the second left intercostal space. The mediastinum was entered transpleurally. A grapefruit sized fatty tumor was found overlying the heart and aorta within the mediastinal pleura. A projection from the main mass extended up through the superior thoracic aperture into the neck (Fig. 2). The tumor which was well encapsulated was removed without difficulty. The lung was re-expanded by means of positive intratracheal pressure and the chest was closed without drainage.

The hospital course was uneventful and the patient was discharged on the ninth postoperative day.

The pathologic report was lipoma.

Follow-up chest films showed expansion of the lung and absence of mediastinum mass density.

DISCUSSION

Although the correct diagnosis was suspected preoperatively in this case, surgery is indicated in all mediastinal tumors. It is the only means of positive

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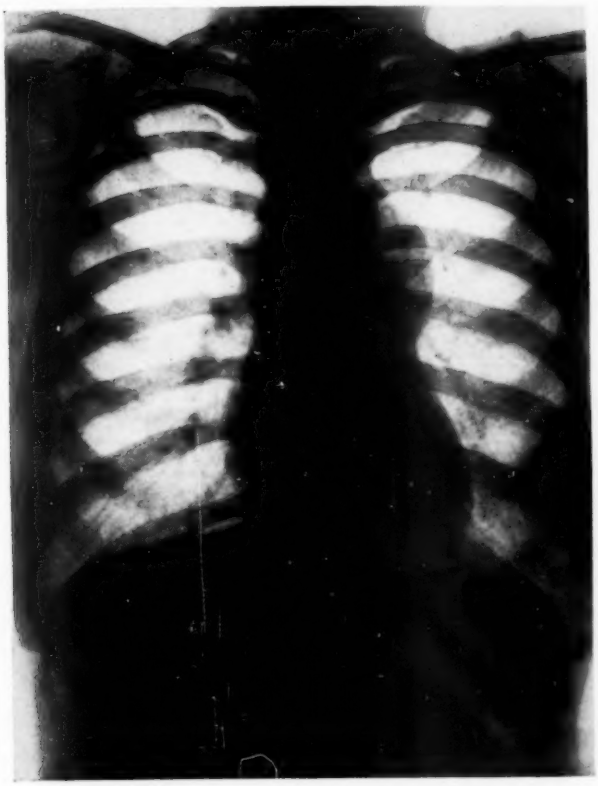

FIG. 1.—Preoperative roentgenogram of the chest.

FIG. 2.—Photograph of tumor (cut surface).

diagnosis in the non-lymphomatous types. Moreover, it is the only definitive therapy. There is futility and danger in giving radiation therapy to mediastinal tumors of unknown nature. The too common practice of employing a "test dose" of deep roentgen ray therapy may erroneously place the label of malignant lymphoma on a patient whose prognosis is, in reality, more favorable. The majority of cases of Boeck's sarcoid show mediastinal node involvement and this often simulates roentgenologically other mediastinal tumors. Many of these shadows show regression or even complete disappearance under observation.[7] Radiation therapy, in this period, would lead to a faulty diagnosis and prognosis. It is equally true that valuable time may be lost with radiotherapy and a once operable tumor become inoperable. Thus a patient's "day of grace may be sinned away" with a modality which is not curative. Pathologic diagnosis should be reached by thoracotomy which, in properly selected cases, is a safe procedure.

In the second place, even if it were known definitely that the tumor was a lipoma, it is probable that pressure symptoms would eventually manifest themselves and make surgery mandatory. That lipomata may attain tremendous size is well illustrated by Heuer's case in which the tumor filled the entire left hemithorax. Death from pressure has been reported.[1, 2] Thirdly, malignant degeneration or infection must be considered as possible complications.

Exploration through the anterior intercostal route without division of ribs gave adequate exposure in this case and, we feel, a more comfortable convalescence to the patient.

CONCLUSIONS

Mediastinal lipoma is a fairly rare disease. The treatment is surgical. The futility of giving radiation therapy to mediastinal tumors of unknown nature is stressed.

SUMMARY

A case is presented in which a lipoma was successfully removed from the anterior superior mediastinum.

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